

Pre- and Postnatal Ultrasound Diagnosis and Maternal Psychological Reactions



Connie Jørgensen

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Title and subtitle Pre- and Postnatal Ultrasound Diagnosis and Maternal Psychological Reactions			
Abstract Ultrasound scannings of pregnant women have become an useful tool in the hands of the obstetrician. At the Department of Obstetrics and Gynecology, University Hospital of Lund, Sweden ultrasound examinations have since October 1980 been used routinely in every pregnancy in the 17th and 32nd week of gestation. The primary intention of the examinations is to obtain a reliable estimation of the pregnancy length and later, during pregnancy, to control the fetal growth. The examinations imply that fetal malformations are discovered. Normal range curves for growth of the fetal cerebral ventricle system during pregnancy were obtained in a material of 654 consecutive pregnancies. These curves were used in a 13 month and a 4 year screening program in order to discover incidences of fetal brain ventricle dilatation. Development during pregnancy, delivery and later outcome of the infants were followed. The normal range curves were used in a study of neonatal acquired ventricle dilatation in 27 infants. For all these infants there were complications in pregnancies, deliveries and/or neonatal period of life. The outcome of the infants was generally very poor with 13/27 not surviving. Poor outcome was correlated to ante- intra- and neonatal asphyxia, seizures, persistent pathological electroencephalogram and assisted ventilation for more than 1 week. In a 24 month period 6020 pregnant women were routine ultrasound examined. Fetuses with malformation of the intestinal or urinary tracts were found in 6 pregnancies. These were followed in order to evaluate whether the prenatal diagnosis had any influence on surgical intervention and prognosis for the child. During a 3 year period 27 cases of fetal malformation were diagnosed by routine ultrasound screening. Semi-structural interviews were made with 24 of the women, 10 had the information after the examination in the 17th week of gestation and 14 after the 32nd week examination. The aim of the studies was to improve understanding of the women's mental reactions. Early diagnosis gives the woman possibility to terminate the pregnancy whereas in late diagnosis the woman has to continue the pregnancy and give birth to a defect child with an uncertain future. The women all experienced a severe psychological trauma, for several longlasting. Prenatal diagnosis of fetal malformations provides a possibility to improve management during pregnancy and delivery as well as earlier postnatal intervention. However, the use of ultrasound is not only a question of developing a satisfactory technique; the psychological consequences must be considered as well.			
Key words Ultrasound, fetal, infants newborn, malformations, prenatal diagnosis, central nervous system, hydrocephalus, brain ventricle dilatation, intestinal and urinary tracts, brain examinations, intracranial hemorrhage, posthemorrhagic hydrocephalus, asphyxia, outcome, neurodevelopment, maternal psychological reactions, selective abortion			
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Connie Jörgensen, Department of Obstetrics and Gynecology
 University Hospital of Lund, S-221 85 Lund, Sweden

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PRE- AND POSTNATAL ULTRASOUND DIAGNOSIS
AND MATERNAL PSYCHOLOGICAL REACTIONS

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Pre- and Postnatal Ultrasound Diagnosis and Maternal Psychological Reactions

Connie Jörgensen



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The thesis is based on the following papers, which will be referred to by their Roman numerals.

- I. C. JÖRGENSEN, I. INGEMARSSON, E. SVALENIUS, S. MONTAN and N.W. SVENNINGSSEN: Ultrasound measurement of the fetal cerebral ventricles - A prospective, consecutive study. Journal of Clinical Ultrasound (accepted 1984)
- II. C. JÖRGENSEN, I. INGEMARSSON and N.W. SVENNINGSSEN: Prenatal diagnosis of fetal brain ventricle dilatation. British Journal of Obstetrics and Gynaecology 90:162-166,1983
- III. C. JÖRGENSEN, I. INGEMARSSON and N.W. SVENNINGSSEN: Brain ventricle dilatation in severely ill newborn. A clinicopathological ultrasonographic study of possible causes in the ante-, intra- and neonatal period. Journal of Perinatal Medicine (submitted 1984)
- IV. C.M. KULLENDORFF, L.T. LARSSON and C. JÖRGENSEN: The advantage of antenatal diagnosis of intestinal and urinary tract malformations. British Journal of Obstetrics and Gynaecology 91:144-147,1984
- V. C. JÖRGENSEN, N. UDDENBERG and I. URSING: Ultrasound diagnosis of fetal malformation in the second trimester. The psychological reactions of the women. Journal of Psychosomatic Obstetrics and Gynaecology (accepted 1984)
- VI. C. JÖRGENSEN, N. UDDENBERG and I. URSING: Diagnosis of fetal malformation in the 32nd week of gestation. A psychological challenge to the woman and the doctor. Journal of Psychosomatic Obstetrics and Gynaecology (accepted 1985)

Abbreviations and Definitions

- AD: Abdominal diameter of the fetus measured as a mean of the anterior-posterior diameter and the transversal diameter. A parameter used for growth control.
- BPD: Biparietal diameter. A parameter used for dating and for control of fetal growth.
- CTG: Cardiotocography. Registration of the fetal heart rate and the uterine contractions.
- GMH: Germinal matrix hemorrhage same as SEH
- EEG: Electroencephalogram
- FL: Femur length. A parameter used for control of fetal growth.
- HW: Hemisphere width in the fetal or neonatal brain measured from the midline structures to the inner table of the skull.
- IVH: Intraventricular hemorrhage
- LVW: Lateral ventricle width measured from the midline structures to the first strong echo from the lateral wall of the anterior horn of the lateral ventricle.
- NICU: Neonatal Intensive Care Unit
- Preterm delivery: Delivery before 36 completed gestational weeks
- SATA: Spatial-average, temporal-average
- SEH: Subependymal hemorrhage same as GMH

Introduction

HISTORICAL BACKGROUND

As long as man has been able to write he has recorded impressions of monster births. One of the earliest of these records is from Chaldaea (2800 B.C.) about abnormal fetuses of the ancient Babylon found on the brick tablets of Koyngik near the Tigris. The Babylonians correlated the birth of a monster to the position of the stars, and the infant was supposed to convey the stellar meaning (44, 77). In the fifth century B.C. the prevailing theories were that variations in the paternal element (Empedocles of Agrigentum in Sicily) or abuses of coitus (Democritus of Abdera) could result in monster infants. None of them ascribed any importance for the reproduction to the mother and regarded her merely as an "incubator". Aristotle (384-322 B.C.) condemned these theories believing that the mother contributed to the fetus and that changes in the female element could lead to malformations. He even rejected the hybrid theory, i.e. that intercourse between human beings and animals could result in monsters. In spite of this, such theories existed until the eighteenth century (44, 77).

During the following centuries grossly malformed infants were regarded as divines and messengers from the Gods. Later it was believed that they were in collusion with the devil and the progress to destroy these infants followed. The Spartans threw their malformed children into an abyss and often the mother did not escape the fate of her child. During the Middle Ages this was still the prevailing belief and many unfortunate women became victims of the flames.

During the 12th and 13th century Albert the Great (1193-1280) and Thomas of Aquinas (1227-1274), both Dominican monks, stated that congenital abnormalities could be caused by faulty material or celestial influence (44, 77, 82).

In the 17th century Henricus Asteldius proclaimed that a Danish astronomer had discovered the origin of monstrosities. He ascribed them to comets which he regarded as tumors scattered throughout the firmament and when precipitated upon earth they could take on all kinds of unusual and extraordinary forms. The moon has been the heavenly body paid most attention to because of the 28 days cycle for both the woman and the moon, e.g. the term "mooncalf" used by Shakespeare in *The Tempest* when referring to Caliban as "a freckled whelp, hag-born - not honoured with a human shape" (44, 77).

During the 16-17th centuries several publications with collections of monsters appeared, e.g. *Lycosthenes* 1557, *Licetus* 1616 and *Aldrovandus* 1642, and flysheets with illustrations of these monsters were scattered among common people (25, 82, 96). During the same period, imagination and fantasies of the pregnant woman were considered decisive for the origin of a malformation. Even the mere sight of certain animals would induce abnormalities such as harelip after regarding a hare or marked growth of the hair after regarding a bear. In 1727, J.A. Blondel concluded in his book "The strength of Imagination in Pregnant Women Examined" that conception, growth and sexual determination of the embryo were all outside the power of the mother's will. Nevertheless, the old belief still existed to the beginning of the 20th century (25, 77, 82).

A more systematic scientific study of fetal malformations started in the 18th century with the work by the embryologist Caspar Friedrich Wolff (1733-1794, who described the wolffian duct). He stated that, formerly regarded as expressions of influences contrary to nature, malformations were explicable by the application of definite laws of development. Around 1820, the German pathologist, Johann Friedrich Meckel (1781-1893, who identified Meckel's diverticulum) published his "Atlas of Human Abnormalities". Nine years later Charles Robert Darwin's (1809-1882) book "The origin of species" appeared. An early experimental work was published in 1826 by the zoologist Geoffroy-St-Hilaire. By different manipulations of chicken eggs he was able to induce a large number of anomalies,

e.g. spina bifida (44, 77, 82).

Parallel with this expansion of teratology it was a popular amusement, still in the 19th century, to collect dwarfs and giants at the courts in Europe, and malformed individuals were exhibits on markets, e.g. the famous conjoined twins Chang and Eng Bunker (from whom the word siamese twins derives). They died in 1874 at 63 years of age. They were rich, married two sisters and had 5 and 6 children, respectively (25, 96, 192).

In the first half of the 20th century there was an increased interest in fetal pathology. Thus, in 1902 Ballantyne from Edinburgh published his "Antenatal Pathology" (81, 189), and in 1906 Ernst Schwalbe (1871-1920) a book on morphology of malformations. In his thesis on "Symmelias and Sirens" in 1914, Paul Barutaut concluded that the etiology was unknown, the pregnancy normal and that the malformations implied the death of the newborns a few hours after delivery (8). In Sweden, the leading teratologist Ivar Broman (1868-1946), professor of anatomy at the University of Lund, considered abnormal ontogeny to be the main cause of malformations (24, 82).

In the last 40-year period a better understanding of the etiology of fetal malformations has developed. In 1941, Gregg showed an increased rate of congenital cataract and heart diseases in children whose mothers had had German measles during the first 3 months of pregnancy (81, 136). The teratogenic effects of toxoplasmosis were described in 1958 (61). In 1952-54 the first reports of the radioactive effects on fetal development from the atomic bomb explosions over Hiroshima and Nagasaki appeared (176, 213). In experimental animals cortisone (Fraser 1954) and vitamin-D deficiency (Kalter and Warkany 1959) were found to be teratogenic with increased frequency of cleft palates in the offspring (70, 113). In 1952, Hsu T.C. and Ford, in 1958, described the karyotype in man (69, 102). In 1959, Jacobs et al demonstrated that mongolism was characterized by having Trisomy-21 chromosome abnormality (105).

Malformations in connection with trisomies 17-18 and 13-15 were described during the years 1960-62 (59, 170). In 1962, the Thalidomid catastrophe happened resulting in a high frequency of amelia and phocomelia as the characteristic injury among the infants (139). In 1961, Fraser arranged the first international conference on congenital malformations.

Several other discoveries and theories, both in our own century and of the past, could be mentioned. However, we are still left with the fact that 75-80% of congenital malformations are caused by an interplay of several genetic and environmental factors. Therefore remains often unanswered the question "Why did this have to happen"?

The era of intrauterine diagnosis started around 1899 when it became possible to demonstrate the fetal head, spine, pelvic bones and long bones by X-ray pictures (37). In 1917, the first scientific report on intrauterine diagnosis of fetal malformations "Anencephaly successfully diagnosed before birth" was published by James T. Case, St. Lukes Hospital, Chicago (37). He found this kind of diagnostics important to avoid Cesarean section in pregnancies with a monstrous fetus and that it could be used safely at all kinds of pregnancy complications.

In 1942, Dussik for the first time made attempts to use uni-dimensional ultrasound as a diagnostic tool for intracranial processes (57). In 1950, Ballentine was able to discover intracranial pathology (5). These investigators together with others started the use of Echo-encephalography for diagnosis of dislocation of the cerebral midline and measurement of the lateral ventricle width, now well established. In the field of echocardiography Edler and Hertz in Sweden were pioneers and described in 1954 a uni-dimensional method for investigation of the movement pattern of some heart structures. (58). In 1952, Howry and Bliss were able to demonstrate a gallbladder containing stones, in vitro using a two-dimensional system (101).

During the last years of the 1950's the two-dimensional ultrasound became the leading method and in 1958 Donald et al introduced the method for investigations in obstetrics and gynecology (54). In 1960, the method was introduced in Sweden by Sundén, described in his thesis in 1964 "On the diagnostic value of ultrasound in obstetrics and gynaecology" (199, 200). Already in 1963, Mac Viae and Donald could demonstrate fetal echoes in the 8th-9th week of gestation (155) and in 1964, Sundén found heart activity in the 13th week of gestation (200). In 1961, the now well known concept BPD (biparietal diameter) was described by Donald and Brown (55). Willock et al demonstrated in 1964 a correlation between BPD and fetal weight (210), and in 1969, Campbell suggested BPD as useful for gestational dating (28), later also proved by Persson et al in 1978 (174). In 1961, the first fetal malformation (hydrocephalus) was diagnosed by ultrasound by Donald and Brown (55). In 1964, Sundén described the intrauterine diagnosis of anencephaly (200). Garrett et al in 1970 for the first time presented the ultrasound image of a polycystic kidney (74). In 1972, Campbell reported the first termination of a pregnancy because of anencephaly diagnosed by ultrasound (29). The normal fetal anatomy was described in detail by Robinson et al already in 1968 and Flanigan et al 1977 (68, 183).

New possibilities of diagnostic ultrasound were introduced with gray-scale and real-time techniques (first used in the Vidoson of Siemens around 1968). The technical equipment has developed rapidly in the 1970's and 1980's implying that the fetus now can be observed in detail during the first part of the second trimester of the pregnancy (Fig 1). In 1981, Stephenson and Weaver in a review of the literature found that around 90 different fetal conditions had been diagnosed by ultrasound and that the amount of papers was overwhelming (197).

The interest concentrated early upon the consequences of the prenatal diagnosis. This is evident from the intrauterine treatment,

already starting in 1963, with Liley's first intrauterine transfusion of blood intraperitoneally to a fetus suffering from Rh-immunization (148). During the 1980's, reports have been published on surgical treatment of fetal malformations, e.g. shunt-operations for hydrocephalus and hydronephrosis (11, 43, 52, 78, 89). This field of surgery started with great enthusiasm. However, since neonatal intensive care has advanced remarkably in later years, premature delivery with extrauterine treatment is preferred in most cases. Thus, intrauterine surgery remains the best choice in a few, very distinctly selected cases only (11, 43).

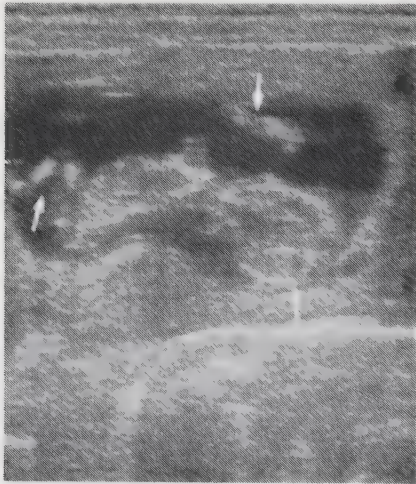


Fig.1. Fetus in the 15th week of gestation. Arrows pointing at fetal head, arm and leg.

Parallel to the progress in obstetric and gynecologic diagnostics the ultrasound became important in most other medical disciplines. In 1979, Pape et al for the first time presented a two-dimensional ultrasound method for brain scanning in newborn infants with the purpose to discover intracranial diseases. They were able to demonstrate hemorrhages in the parenchyma and in the ventricles, ventricle dilatation and cerebral infarctions, and they concluded that this was a safe and simple non-invasive technique (165). Previously computerized tomography (CT) had been the only way to diagnose such changes in infants. Comparisons between CT and two-dimensional ultrasound showed that in most cases the ultrasound findings were in accordance with those of CT and autopsy (79, 154, 163, 166, 193). Serial ultrasound examinations showed that intracranial hemorrhage occurred much more frequently than previously

known, especially in preterm infants (9, 141, 169, 198).

Not only hemorrhages but other pathological intracranial findings became also possible to investigate (145, 173, 195). The most distinctly recognized pathological change is dilatation or enlargement of the lateral brain ventricles. One major cause is an intraventricular hemorrhage with distension of the ventricles (80, 93, 97, 143, 187), but cerebral atrophy with thinning out of the cortex may also cause enlarged ventricles (92, 198).

The intracranial findings have been correlated to later outcome (2, 92, 93, 206, 211). Several systems for grading the extent of the hemorrhages have been developed with subsequent difficulties in comparing the results (2, 45, 144, 158, 167, 191). The longterm outcome of the infants is still uncertain although large hemorrhages usually imply a high risk of future handicaps (92, 198, 211). However, a uniform system for grading of the hemorrhage and a systematical evaluation of the corresponding outcome are still missing.

Ultrasound scanning of the skull has now developed into an established supplement to other clinical examinations of the newborn infant. It can be used at the bedside even in severely ill infants with minimal disturbance of the infant and the nursing care.

The consequences for women, giving birth to a malformed baby, have varied considerably during the history of man, from being come upon by Gods or allied with the devil, to having intercourse with animals or being influenced by mental impressions. It was not until our own century that it was stated that the mother is without guilt when having a baby with congenital malformation, since the cause is now considered to be a combination of several genetic and environmental factors. Guilt and feelings of inferiority might have been common among these women, but literature supporting this is sparse.

In 1933, the psychology of female sexuality and reproduction was

emphasized by Horney and later by Deutch (1945) and Benedek (1952) (10, 53, 99). In 1959 and 1961, Bibring described psychological changes during pregnancy (14, 15) and in 1963, Brazelton pointed out the importance of early mother-infant adjustment (22). The special psychological problems at premature birth have been studied by Caplan (1960), (35). In 1968, D'Arcy and Daniels both reported the psychological reactions seen in families getting a child with a congenital malformation (48, 49). Many papers have dealt with psychological reactions in connection with still-birth, early neonatal loss and legal abortion (46, 116, 119, 131, 137). Lately, papers concerning pregnancy termination after amniocentesis (19, 140) and women's reaction to ultrasound examinations have appeared (178, 179). However, psychological reactions experienced by women at an unexpected discovery of a fetal malformation during routine ultrasound examination have not previously been described. The crisis in connection with a psychological trauma has been described by Cullberg (1978), and is characterized by four stages: shock-phase, reactionphase, phase of working through the trauma and re-orientation (47).

Present Investigation

The application of the ultrasound technique has got a prominent position in obstetrics. Fetal malformations still present diagnostic difficulties, especially in early pregnancy. Knowledge of normal fetal organ development, the origin of early or late diagnosed malformation and the implications for the child are necessary prerequisites for utilizing ultrasound in prenatal diagnosis. We therefore decided to study in detail fetal intracranial anatomy, especially the brain ventricles. Firstly, it was necessary to follow the normal growth of the brain ventricles during intra-uterine life (I). Secondly, the result of the investigation was used to evaluate the frequency of abnormal findings in a routine ultrasound screening of a pregnant population and was related to the outcome in the infants (II). Ultrasound brain-scanning of

newborn infants was introduced in the NICU in Lund already in 1980 by the author as one of the first in Sweden. The antenatal and postnatal development of the brain ventricle system in severely ill newborn infants was compared with the normal material (I) in a clinico-pathological study evaluating these data with outcome in the infants (III).

Most fetal malformations diagnosed during the third trimester are not possible to treat during intrauterine life, but a careful follow-up during pregnancy, electively planned delivery and early postnatal treatment may improve the prognosis for some of the children (IV).

The use of ultrasound scannings as a routine during pregnancy implies discovery of unexpected fetal malformations. Unprepared and perhaps even without wanting it the women receive this information. Naturally these circumstances will imply a heavy burden on every pregnant woman and the obstetrician must be prepared to take care of and support the women emotionally (V and VI).

Aims of the Investigation

Part One

To achieve normal values for the growth of the fetal lateral brain ventricles in order to improve the intrauterine diagnosis of ventricle dilatation.

Part Two

To use these parameters in an antenatal screening of a population where also the pathological diagnosis, follow-up during pregnancy, delivery and the outcome of the infants are discussed. To use the parameters in a study of neonatal brain ventricle dilatation where possible causes in the ante- intra- and neonatal period are analyzed in relation to later outcome.

Part Three

To evaluate prenatal diagnosis of fetal malformation during the third trimester.

Part Four

To study the psychological reactions in women receiving information about fetal malformation after an ultrasound scanning during the second trimester (still with the possibility to terminate the pregnancy) or during the third trimester (where the diagnosis implies delivery of a malformed child).

Methods

Technical equipment

The routine ultrasound examinations in Paper I, II and IV were made with a real-time ultrasound scanner Hitachi EBU 22 and 25 (Sonotron) calibrated to the velocity of sound 1540 m/s employing a 3.5 MHz linear transducer. For documentation polaroid photographs were used and in some cases even videotape recordings were made.

The 27 newborn infants in Paper III were examined with one of the linear scanners or with a sector scanner, Diasonic DS 20 calibrated to the sound velocity 1540 m/s employing a 3.5 MHz transducer or a special 6.0 MHz neonate transducer. The ultrasound intensity was $5,7 \text{ mW/cm}^2$ and 7 mW/cm^2 (SATA) for the linear scanners, respectively and $6,9 \text{ mW/cm}^2$ (SATA) for the sector scanner (according to the manufacturer).

Method for measurement of fetal lateral brain ventricles and ventricular/brain index.

Fetal biparietal diameter (BPD) was measured on the broadest oval section of the fetal head with a clear image of the midline structures, from the outer to the inner table of the skull (Campbell 1968 and Persson et al 1978), (27,174). At the same level, or by moving the transducer in cranial direction, the anterior horns of the lateral brain ventricles appear as two short lines parallel to the midline echo. At the same level even the posterior horns may be demonstrated (Fig. 2). Cranial to these structures the midbody of the ventricles can be found as two centrally placed lines parallel to the midline echo (Fig. 3).

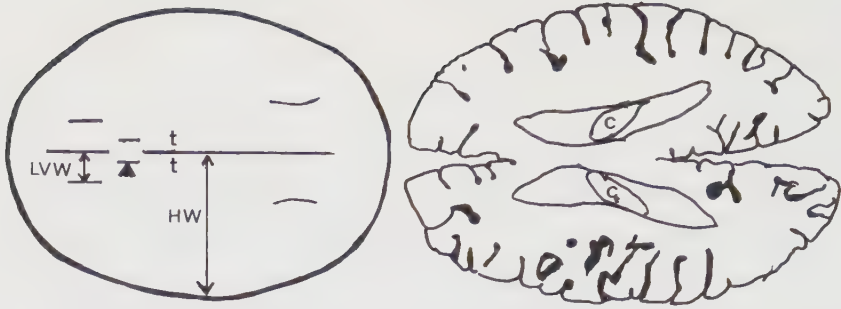


Fig 2. Drawings: Left illustrating the ultrasound scan through the level of the frontal horns of the lateral ventricles. LVW = lateral ventricle width, HW = hemisphere width and arrow pointing at cavum septi pellucidum. Right the corresponding brain cut.

t = thalamus, c = chorioidal plexus



Fig 3. Drawings: Left illustrating the ultrasound scan through the midbody of the lateral ventricles. Right the corresponding brain cut.

We found the visualization of the fetal anterior horns to be most accurate and reproducible. In Paper I the width of the fetal lateral ventricle (LVW) was measured from the midline structure to the first strong echo of the lateral wall of the anterior horn. At the same level the width of the hemisphere (HW) was measured from the middle of the midline echo to the first strong echo of the inner table of the skull (Fig. 2). The parameters were measured on both sides and the results are given as the mean values. In Paper I LVW

and HW were measured according to gestational age and BPD, the gestational age being established from measurement of the BPD in relation to a normal range curve (Persson et al, 1978). From the values of LW and HW the ventricular index LW/HW could be calculated.

In Paper II measurements of the midbody of the lateral ventricles as well as the anterior horns were used. We found that it was easier and more reliable to use measurement of the midbody instead of the anterior horns in cases of pronounced ventricle dilatation. The LW midbody was measured in the same way as described in Paper I, i.e. from the middle of the mideocho to the first strong echo of the lateral wall of the ventricle, and the hemisphere from the middle of the mideocho to the inner table of the skull (Fig. 3). From LW midbody and HW the ventricular index was calculated. This method is well described by Denkhaus and Winsberg (1979), Johnsson et al (1980), Jeanty et al (1981) and Hadlock et al (1981) giving normal values according to gestational week (menstrual age) (51, 85, 106, 109), and by Fiske et al (1981) for diagnosis of early ventricle dilatation (65).

Methods for measurement of the neonatal lateral ventricular width and the LW/HW-index.

In Paper III a 3.5 MHz linear transducer was used and later even a sectorscanner with a 3.5 MHz or a 6.0 MHz neonate transducer was available. The linear transducer was placed against the temporo-parietal region of the infant's head at the level of the outer canthus of the eye and the helix of the ear. Kept parallel to this line the transducer was moved cranially until the body of the lateral ventricles was clearly demonstrated (Fig. 4 left). The distance from the midline echo to the lateral wall of the ventricle was measured on the contralateral side which is always seen most clearly. At the same level the hemisphere width was measured as previously described. For measurement of the other ventricle the transducer was placed in the same way on the other side of the

neonatal head.

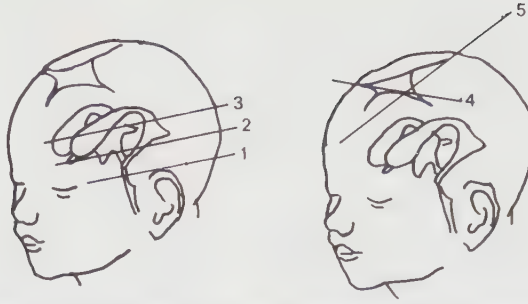


Fig 4. Drawings illustrating the systematic ultrasound examination of the neonatal skull. Left Transversal scans 1: a level at the base of the brain, 2: a level through the third ventricle and 3: a level through the midbody of the lateral ventricles. Right 4: coronal scan and 5: sagittal scan.

The same points of reference were used with the sector scanner most suitable for transfontanel scannings. Both linear and sector scanner transducers were placed over the fontanel to produce supplementary coronal and sagittal scannings (Fig.4 right) with the purpose to diagnose intracranial hemorrhages and atrophy .

All neonatal scannings were performed by one person (CJ) bedside at the Neonatal Intensive Care Unit (NICU) (Fig 5).



Fig 5. Skull ultrasound examination in a preterm infant.

The ultrasound findings in Paper II and IV were compared to the neonatal findings in order to investigate the accuracy of the prenatal diagnosis.

In Paper III the hospital files of mothers and children were investigated for ante- intra- and neonatal factors of importance for the occurrence of brain ventricle dilatation and later outcome in severely ill newborn babies. The neurodevelopmental stage was evaluated at 1 1/2 - 3 years of age according to Milani-Comparetti developmental score (160).

Interview methods

Paper V and VI are based on interviews with 10 and 14 women respectively who, after a routine ultrasound scanning, were informed that their fetuses were malformed. The interviews were semi-structural in Paper V and modified semi-structural in Paper VI, implying that previously decided answer-alternatives were not used in Paper VI for categorization of the answers. In Paper V 21 items were discussed (see questionnaire in appendix page 54) in Paper VI 20 items (see questionnaire in appendix page 56). The questions in Paper V concerned the information, the decision for termination of the pregnancy and the psychological reactions. In Paper VI the information, the remaining part of the pregnancy, the delivery and the time that followed were discussed. The time interval between pregnancy and interview varied from 7 months to almost 3 years.

Statistical methods

In Paper I the data were computeranalyzed resulting in mean value, standard deviation (SD) and standard error of the mean (SEM) for LVW, HW and LVW/HW-index according to BPD and gestational age. Significance test in Paper I was the Student-T test. In Paper III the χ^2 -test was used and in Paper VI Fishers exact test.

Methodological considerations

Our normal curves for growth of the fetal brain parameters LVW and HW were constructed from measurement of the anterior horns of the lateral ventricles. In the study of ventricle dilatation in Paper II and III we used measurement of the midbody of the lateral ventricle. A comparison between these two methods may imply errors. Fig. 6 shows a graphical comparison between our results and the results of Denkhaus and Winsberg, Johnson et al and Jeanty et al, as the index LVW/HW against BPD. Denkhaus and Winsberg used the same method as we did but reported their result as bi-anterior horns size and BPD instead of hemisphere width and gave the index as LVW/BPD against BPD. Johnsson et al and Jeanty et al used the midbody measurement, their results are given as LVW midbody/HW against gestational age from the last menstruation and not gestational age calculated from ultrasound as used in this investigation. In Fig. 6 we have transformed gestational weeks to BPD values using the normal curve of Persson et al (174). In spite of these methodological differences it is seen that the shape of the curves is approximately the same. We found a SD of 6 mm in gestational week 17 and 3.5 mm from week 30 to term. With this in mind, the error made by using the standard curves from Paper I for evaluation of prenatal and neonatal ventricular dilatation in Paper II and III respectively, can be considered of minor importance.

In Paper III we used our intrauterine standard parameters for evaluation of neonatal ventricle dilatation. We therefore compared our results to those of Levene (142) from newborn infants born at the age of 28 to 42 gestational weeks (Fig. 7). We found a linear growth of LVW against gestational week whereas Levene found a wave form. Still, the differences between our results and those of Levene are less than one SD. The reason for the different shape of the curves could possibly be changes in cerebral blood flow and compression of the neonatal head, which is probably more pronounced in extrauterine than intrauterine life (142).

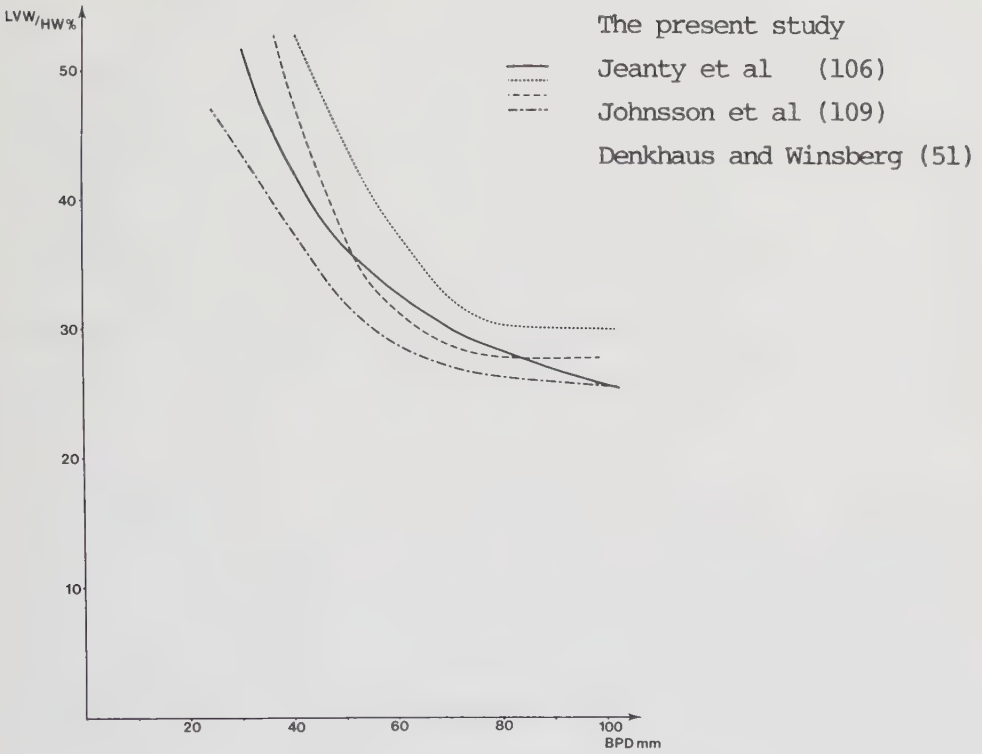


Fig 6. A comparison between the results in the present study and three other reports. LVW/HW given in % against BPD in mm.

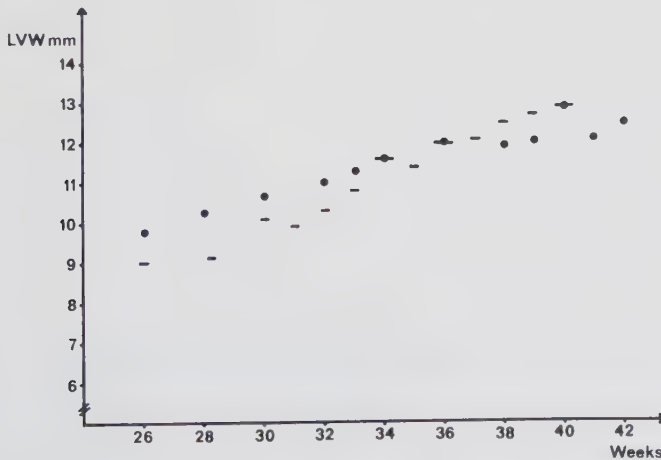


Fig 7. A comparison of the growth of LVW mm against gestational age in weeks (Paper I ■, Levene 1981-).

Semi-structural interviews may limit the woman's spontaneous description of her experiences, the interviewer interrupting with new questions. We were aware of this and tried to avoid it by actively choosing questions fitting best to the thoughts and expressions of the woman without considering the order in the questionnaire. The studies were retrospective, implying that a standardized time from pregnancy to interview could not be used. Also the fact that the interviewer in most cases had been responsible for the diagnosis and obstetrical management may have influenced the answers. The results in Paper V and VI give preliminary impressions valuable to the clinician and for designing a future prospective study.

Patients

Ultrasound scanings have been used at the Department of Obstetrics and Gynecology, University Hospital of Lund, as a diagnostic tool since 1960. In October 1980, the program of routine ultrasound scanning started. Every pregnant woman is offered an examination in the 17th and 32nd week of gestation. At the early examination the size of the fetus is measured (BPD, AD, FL) and the gestational age established, the number of fetuses is registered and information of any fetal abnormality is obtained. At the 32nd week examination the primary intention is to control fetal growth and to locate the placenta. Even at this examination fetal malformations may be discovered. The routine screenings are performed by specially trained midwives under supervision of a doctor. The investigations of this thesis are based on these routine screenings, except for Paper III.

Paper I includes in total 654 consecutive pregnancies. Routine ultrasound examinations were made in the 17th and 32nd week of gestation. On a voluntary basis, after verbal information, additional scanings were made in 408 patients in week 39 (38-40) and after term once a week. The re-examinations were of no consequences

for most of the women, but for some signs of fetal distress (i.e. decreased growth since the 32nd week examination, extreme oligo-hydramnios or decreased fetal movement) made obstetrical intervention necessary. In Paper I we have only a few examinations between week 17 and 32, since fetal growth in this period has been well documented by others (51, 106, 109).

Paper II comprises 3125 women examined consecutively by routine ultrasound during a 13 month period. Fetuses with dilatation of the brain ventricles were observed in 5 pregnancies. The results are presented in the paper as case reports concentrating on fetal development, obstetrical management and outcome of the child.

Paper III comprises 27 newborn infants developing dilatation of the lateral brain ventricles. They were all treated in the Neonatal Intensive Care Unit (NICU) because of complicated pregnancies and/or deliveries. The dilatation of the lateral brain ventricles was in 20 of 27 cases probably due to intraventricular hemorrhage (IVH). The occurrence of dilated ventricles at ultrasound was used as the selection criterion.

The total number of neonatal scannings performed during the same period was 412 in 166 infants. The study of Paper III was made in order to analyse the importance of various factors for the development of brain ventricle dilatations in newborns with perinatal complications.

Paper IV includes the total population of 6020 routinely examined pregnant women during a two-year period. Fetuses with congenital malformation of the intestinal or urinary tracts were found in 6 pregnancies. These 6 pregnancies were followed in order to evaluate whether the prenatal diagnosis had any influence on surgical intervention and the prognosis of the child.

Paper V and VI During a three-year period, 27 fetal malformations were diagnosed in 8422 women, examined with routine ultrasound.

These women were contacted by means of an informative letter after permission from the Local Ethical Committee. Later, a telephone call was made where additional information was given and arrangements for the interview were made. The participation was voluntary. However, all women expressed interest for the aim of the study and were willing to partake.

Two of the 27 women did not participate in the interview study. One had left Sweden for Zimbabwe, and one woman had an intrauterine death in the 36th week of gestation. Her fetus had an extremely dilated urinary bladder, possibly a defective neuromuscular transmission of unknown etiology. This woman was excluded from the study as this was the only antenatal death. A third woman was pregnant with a malformed fetus for the second time.

Paper V comprises interviews with 10 women who received information about a fetal malformation discovered at routine ultrasound scanning in the second trimester, and Paper VI interviews with 14 women informed in the third trimester.

These results are reported separately because the situation for these two groups of women was very unlike. The first group (Paper V) made an active decision to terminate the pregnancy. The second group (Paper VI) had a malformed child for whom they could neither interfere nor change the destiny. The results also show that these women had quite different experiences.

Results and Comment

Part One

Measurement of the fetal lateral brain ventricles.

Fetal neuroanatomy became early an area of interest for investigations of fetal malformations. The reason is probably that fetal

biparietal diameter (BPD) was used for estimation of fetal growth since 1961 and since 1969 for dating of pregnancy. Finally, a neural tube anomaly is a fairly common malformation in human fetuses. In 1977, Campbell described a method for investigation of the fetal spine already in the 16th-19th week of gestation (33, 34). He also showed that enlargement of the lateral ventricles above 50% of the hemisphere width is characteristic for concomitant hydrocephalus. Analysis of maternal serum alfa-fetoprotein (AFP) for diagnosis of neural tube defects was introduced by Brock and Sutcliffe in 1972 (23). In his 1977 study of AFP screening in 308 pregnancies with increased risk of neural tube defects, Campbell found 28 with high AFP and neural tube defects. Another 4 pregnant women with raised amniotic AFP had, however, normal fetal ultrasound examinations and later delivered a normal child. Finally 7 pregnant women had raised AFP due to a gestational age being 3 or more weeks shorter than predicted from last menstruation. With ultrasound scanning 25 of 28 cases of neural tube defect could be diagnosed and thus a false positive diagnosis be avoided. In 1983, Persson et al found raised AFP in serum in 1,2% of a normal population. There were two false negative AFP-tests in serum in the screened population of 10147 pregnancies. Amniocentesis in 117 of the 120 pregnancies with elevated serum AFP revealed 9 cases with raised values of which 2 had a normal ultrasound examination and delivered a normal child. These authors concluded, as Campbell did, that AFP-screening may be valuable when combined with ultrasound examinations for both pregnancy dating and for excluding false positive cases (175).

Already in 1977, Campbell reported that BPD in fetuses with spina bifida often is smaller than the average for gestational age (34). This was confirmed by Wald et al in 1980 and Chervenak et al in 1983 (41, 203). Thus, only by measurement of the fetal brain ventricles will it be possible to diagnose a concomitant hydrocephalus due to the Arnold-Chiari malformation. Even hydrocephalus without spina bifida may have a normal BPD, at least during the first part

of the pregnancy. Ventricle dilatation may precede cranial enlargement by several weeks (34, 95, 132).

Paper I deals with results of measurement of the anterior horns of the lateral brain ventricles in 654 consecutive pregnancies. All pregnancies were dated with BPD measurements according to the normal range curve of Persson et al (172). In a few cases it was not possible to visualize and measure the BPD due to the position of the fetus. In the present study the success rate of measurement of the width of the lateral ventricle (LVW) was 90% in fetuses with BPD between 26mm and 103mm as compared to 80% reported in 1980 by Johnson et al (109). We found that the LVW growth versus BPD followed a linear regression line (fig 3 page 84). Denkhaus and Winsberg (1979) reported similar results using bi-frontal horn width against BPD (51). We therefore consider the regression to be linear even in the BPD interval from 50mm to 76mm although we have no observations of our own in this range owing to the design of the study. Further support for this assumption are the findings of Fiske and Filly who in 1982 showed that besides increased cortical convolutions the brain undergoes only minor structural changes after gestational week 24 (67). The growth of the hemisphere width (HW) against BPD also followed, as expected, a linear regression. From LVW and HW the LVW/HW-index was calculated. As shown in Fig 4 our results are in accordance with previous observations (51, 85, 106, 109). In conclusion, the present observations indicate for normal LVW/HW-index an upper limit of 50% in gestational week 17 and 30% after gestational week 32.

In the present investigation (Paper I) we have followed a consecutive series of unselected and not a selected normal series of pregnancies. Therefore we have evaluated whether this could influence the results. Comparisons in relation to birth weight at term showed only differences in size of LVW in gestational week 32 and 39 in infants with birth weight below 3000g and more than 4500g. A comparison of preterm infants and a group of growth retarded term infants, with birth weights in both groups below 2800g,

showed no difference in LVW size in the same gestational weeks. Thus, the growth of LVW is related to gestational age and not birth weight. This is in agreement with the observation of Eik-Ness et al in 1980 that birth weight is related to abdominal diameter (AD) rather than to BPD (60).

By comparison the mean values of BPD and LVW at gestational week 17 and 32 were significantly larger in boys than in girls ($p < 0,01$). The same tendency was found in week 39 and postterm but no significant difference was found in these late gestational weeks, probably due to a smaller number of observations.

Part Two

Fetal brain ventricle dilatation

Fetal brain ventricular dilatation is a heterogen condition. Obstruction to the cerebrospinal fluid (CSF) drainage in the ventricular system causes a non-communicating hydrocephalus and obstruction in the subarachnoidal space a communicating hydrocephalus. Sometimes the increased ventricular size is caused by a cerebral atrophy with parenchymal reduction. Furthermore, in a fetus with myelomeningocele the BPD is normal or even smaller than the mean BPD for the gestational week as shown in Fig 8.

The incidence of hydrocephalus varies in different countries. Carter reported an incidence of 0.25% of myelomeningocele in North and West England. As 80% of the infants usually have concomitant hydrocephalus this diagnosis accounts for 0.20%, compared to the total estimated incidence of hydrocephalus 0,14% in a Swedish postnatal population (36, 123, 152). In our population (Paper II) the rate of fetal brain ventricle dilatation was 0,16%. In this study the ventricle dilatation was diagnosed early in one case with extreme polyhydramnios leading to a legal termination in the 23rd week of gestation. The fetus had a malformed skull and dwarfism with very short extremities. Another 4 fetuses were followed during

the pregnancy with measurement of the LVW/HW-index. None showed signs of increased intracranial pressure with accelerated growth of BPD. Two fetuses had concomitant spina bifida, one a large frontal encephalocele and one infant had no other anomalies. Two of these 4 infants were born vaginally and two by Cesarian section because of a big frontal encephalocele in one case and for psychological reasons in the other. At 6 months to 1 1/2 years of age two infants had slight psychomotor retardation but two had normal psychomotor development.

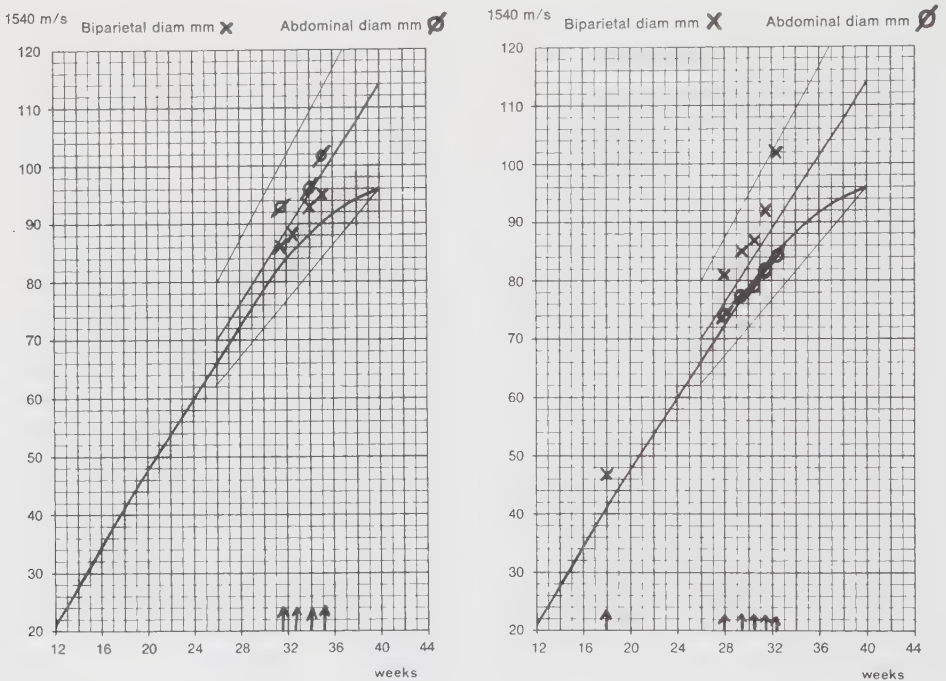


Fig 8. Growth of BPD and AD. Left in a case of myelomeningocele and right in a case of aqueductal stenosis.

Incidences calculated from a 13 months material may not be representative. Therefore the results of the 4-year period October 1980 - 1984, including the studied population in Paper II, obtained by routine ultrasound screening of 11146 pregnancies are presented. Thirteen cases of fetal brain ventricle dilatation were diagnosed i.e. a 0,12% rate. In our follow-up study there was no case of

later infantile hydrocephalus from prenatal causes, i.e. no negative ultrasound scannings during pregnancy. Table I shows the diagnoses of these cases. Five (5/13) cases were diagnosed early with termination of the pregnancy and the diagnosis verified at autopsy. One of these autopsies showed a disturbed cellular migration which later would have caused cerebral atrophy. Extracranial anomalies associated with brain ventricle dilatation occurred in 8 of 13 (60%). Chervenak et al reported a comparable incidence of 63% in a series of 30 fetuses, with chromosomal anomalies in 11% (41).

Table I. Diagnosis of 13 cases of intrauterine ventricular dilatation in a routine ultrasound screened population of 11146 pregnancies.

	Diagnosis in gestational week	
	17	32
Hydrocephalus and spina bifida	3	4
Hydrocephalus due to infection		
Coxsackie-B virus and Toxoplasmosis		2
Meningoencephalocele		1
Dandy-Walker syndrome		1
Thanatophor dwarfism	1	
Disturbed cerebral development	1	
	5	8

Four of 7 infants with late diagnosis were born by CS, the indication being fetopelvic disproportion in one and polyhydramnios with fetal distress in another. As earlier mentioned, the indications for CS in the remaining two were a large frontal encephalocele and psychological reasons, respectively.

The over all outcome in this four year material was legal termination in 5 (38%), 1 death (8%), (this fetus had a trisomy 18) and 7 survivors (54%). In comparison, Chervenak et al (1984) reported 50 cases of brain ventricle dilatation with 26% electively aborted, 46% neonatal death and 28% survivors (42). The outcome of the surviving infants in our material ranges from shunt operation with normal development to severe psychomotor retardation.

One pregnant woman having a fetus with pronounced dilatation of the ventricles was referred to our Department of Obstetrics at gestational week 25. The pregnancy was followed as described. The BPD was above average and accelerating further after week 32 (Fig 8 right). Cesarean section was performed in the 33rd week of gestation and the baby was operated in the Department of Neurosurgery because of aqueductal stenosis with a ventriculo-peritoneal shunt. At 1 year of age this baby has only a slight psychomotor retardation.

A new approach to intrauterine hydrocephalus was presented in 1982 by the Denver group (Clewell et al) who made the first intrauterine shuntoperation (43). Birnholz and Frigoletto had reported their results of repeated cephelocentesis in 1981 (16). From the available results Redwine and Peters (1983) concluded that intrauterine shunting can be valuable only in cases of fetal pulmonary immaturity and hydrocephalus with rapidly accelerating BPD, without structural or chromosomal anomaly or signs of intrauterine infections (181). None of the infants in our four year material fulfilled these criteria.

Neonatal brain ventricle dilatation.

Intracranial hemorrhage is a major threat to the preterm newborn infant and accounts for a great part of perinatal mortality and morbidity (Westgren et al, 1982), (208). In several centers routine ultrasound scannings have been performed in preterm infants finding a high incidence of intracranial hemorrhages especially in infants

with birth weight below 1500g being 51% in the study of Cooke (1981), 43% in the study of Levene et al (1981) and 50% in Stewart et al's study (1983), (45, 141, 198).

The anatomical background for intracranial hemorrhage has been described by Wiggleworth et al and Volpe et al (206, 209). In the germinal matrix (GM) or ependyma in the lateral wall of the lateral ventricles a rete of fine vessels with little supportive connective tissue is present in the fetus from the 28th to the 34th week of gestation. This area is very vulnerable to changes in cerebral blood flow and pressure. Lou et al (153) showed in newborn infants that in asphyxia the autoregulation of the cerebral blood flow (CBF) is impaired. Hypertension or sudden changes in blood pressure are considered the most important causative factors of intracranial hemorrhage in the preterm infant. Asphyxia may cause hypotension with cerebral ischaemia from a reduced cerebral blood flow. When cerebral blood flow is restituted the fragile damaged vessels in the GM-layer may burst and cause a GM or subependymal hemorrhage (206, 209).

In preterm infants the germinal matrix hemorrhages (GMH) or subependymal hemorrhages (SEH) are most common. These may also extend into the ventricular system (IVH) and occasionally occlude it causing a ventricle dilatation. In some infants the hemorrhage may even extend into the parenchyma. In full term infants the same type of IVH can be seen but more often starting with bleedings in the chorioidial plexus (66, 141).

Various systems for grading the extent of the hemorrhages have been proposed and also correlated to the longterm outcome. As shown in Table II, several different ultrasound grading systems exist which differ from those used in CT. Thus comparison between different studies of outcome against degree of hemorrhage is difficult.

According to Hill and Volpe 1981 (45, 93, 187, 198) ventricle

Table II. Grading systems for intracranial hemorrhage by CT and Ultrasound (US) examinations.

References	Technique	Grading system and definitions
Papile et al	CT	1 Isolated SEH 2 IVH without ventricle dilatation 3 IVH with ventricle dilatation 4 IVH with parenchymal extension
Mantovani et al	CT	1 SEH or IVH filling < 10% of the ventricles 2 IVH filling from 10%-50% of the ventricles 3 IVH filling > 50% of the ventr.
Bejar et al	US	Major: SEH + IVH + ventr. dilatation Minor: SEH + IVH without ventr. dil.
Shankaran et al	US	Mild: SEH and or a small IVH Moderate: Intermediate IVH Severe: Total filling the ventricles and parenchymal extension
Levene et al	US	1 SEH with or without inferior or lateral extension of blood beyond most lateral border of ventricles 2 downward extension into the basal nuclei on at least one side or involvement of caudate to genu of ventricle posteriorly on parasagittal scan

- 3 Large hemorrhage with any degree of extension laterally or superiorly into cerebral parenchyma

Allan et al

US

- 1 GMH
 - 2A GMH + IVH filling < 50% of either ventricle
 - 2B GMH + IVH filling > 50% of either ventricle
 - 3 As 2B + intraparenchymal hemorrh
 - 4 Severe IVH filling both lateral ventricles with marked dilatation and large intraparenchymal hemorrhage shifting the midline
-

dilatation occurs in the majority of severe IVH. However, enlargement of the ventricles will occur also in cases of cerebral atrophy. The development of dilatation can be followed during pregnancy with measurement of the ventricle/hemisphere-index as described in Paper I.

Paper III is an investigation of 27 infants who during the neonatal period developed dilatation of the lateral ventricles as diagnosed by ultrasound scannings. An LVW/HW-index of average for the gestational age + 2SD or more was used as selection criterion. Twenty-five of these 27 infants had been examined with ultrasound during intrauterine life. These prenatal examinations showed no signs of congenital ventricle dilatation. Previous studies of the reliability of diagnostic ultrasound versus CT and autopsy found a good correlation in most cases (4, 79, 108, 154, 163, 166, 173, 187, 193). Our results were similar in spite of long time intervals between these examinations in some cases (see Table II appendix page 52). Besides ultrasound, CT, cytology of cerebrospinal fluid and autopsy findings this Table also presents major clinical data

of each baby. In 20 of 27 infants the ventricle dilatation was probably post-hemorrhagic. In seven infants ultrasound showed only a ventricle dilatation. In three of these other disorders were diagnosed i.e. dystrophia myotonica, polyneuropathy and cerebral dysfunction of unknown etiology. In the remaining four infants cerebral atrophy was suspected and later verified by autopsy.

The main purpose of the study (Paper III) was to evaluate in a selected group of severely ill infants some perinatal factors supposed to be important for development of brain damage. The outcome of these 27 infants was very poor since 13 died, 13 survived but with neurodevelopmental handicap and only 1 infant had a normal development at one year of age. The outcome was not related to gestational age. Postnatal mortality was directly correlated to Apgar score <4 at 1 minute and <7 at 5 minutes. A similar correlation was found by Graziani et al (1980), Hill and Volpe (1981) and Tejani et al (1984) but not by Beverley et al (1984), (13, 80, 92, 202). In our material ante/intrapartum asphyxia evaluated from CTG was not significantly correlated to neurological outcome. However, several were emergency CS preterm deliveries (e.g. abruptio placentae) with only a few CTG registrations. Signs of asphyxia in CTG of preterm infants are also much more difficult to interpret than in term infants (Westgren et al), (208). Still, our data indicate that perinatal asphyxia remains an important perinatal factor for development of intracranial disturbances. Other significant factors were respirator treatment (more than 1 week), prolonged seizures and persistent pathological EEG. Clinical seizures were not seen as often in preterm below 30 weeks as in term infants. Preterm and asphyxiated infants may have subclinical convulsions, so-called "silent seizures", with a typical pattern of ictal activity on EEG with continuous cerebral function monitor (Hellström-Westas et al, 1985), (90). A major cause of seizures after intrauterine asphyxia is hypoxic-ischaemic encephalopathy (64). This is correlated to poor outcome as evident also in the extremely preterm infants with brain ventricle dilatation in our study. Such infants with asphyxia had a poor outcome compared to those without asphyxia. Thus, in

the group with asphyxia 5 infants died and 3 had CP at follow-up. Since only 6 infants were born vaginally outcome versus mode of delivery could not be evaluated. Seven of the 27 severely ill infants developed a progressive hydrocephalus needing decompression with a shunt-operation.

Part Three

Prenatal diagnosis of intestinal and urinary tract malformations.

Today many congenital malformations can be corrected by surgical treatment (Hobbins et al, 1979 and Canty et al, 1982), (34, 95) which is important to take into consideration in an evaluation of benefits and hazards of prenatal ultrasound diagnosis. Some malformations may occur with others, sometimes more serious defects (e.g. congenital heart anomalies), which should be taken into account as well.

In 1970, Garrett et al discovered prenatally a fetus with polycystic kidneys (74). The fetal kidney can be visualized by ultrasound from the 15th week of gestation but a diagnosis of malformation is not reliable before the 20th week of gestation (84, 138). In 1973, Campbell and Wladimiroff described a method for measuring the fetal urine production by ultrasound (30).

Experimental urethra obstruction in the sheep kidney in the first half of pregnancy will produce severe renal dysplasia. Later onset is also followed by impaired renal function but to a less extent (8). Therefore Golbus et al 1982 (78), Harrison et al 1982 (88) and Berkowitz et al 1983 (11) have advocated intrauterine decompression with a renalpelvic-amniotic shunt in cases with bilateral obstructions in mid-gestation, but preterm delivery after a gestational age of 33-34 weeks. Unilateral obstructions in general have a good prognosis whereas bilateral obstructions are often accompanied by oligohydramnios and pulmonary hypoplasia as well. In these cases prenatal diagnosis may still be beneficial by allowing early

postnatal examination and treatment.

In our material (Paper IV) two infants had obstructive nephropathies requiring surgical intervention. One infant with distal ureteric stenosis was operated successfully resulting in normalised renal function and one twin baby with unilateral hydronephrosis from obstruction in the uretero-pelvic junction was operated initially with pelviplasty but later nephrectomy was needed. The other twin had an similar malformation but no treatment was required. This baby is not included in the study.

A third infant had a unilateral multicystic non-functioning kidney. When bilateral, such congenital defects (Potter II) are not compatible with extrauterine life. This condition, as well as infantile polycystic disease (Potter I), can be diagnosed early in pregnancy with the possibility to terminate the pregnancy by abortion (84, 112, 115, 132, 184). On the contrary a unilateral lesion has a good prognosis and is in general not combined with other malformations.

The characteristic ultrasound appearance of duodenal atresia, the "double bubble", i.e. two spaces distended by fluid in the fetal abdomen from distension of the stomach and upper part of the duodenum (100), was found in one child in our study (Paper IV). By early recognition and postnatal operation dehydration and electrolyte disturbances were avoided. In another infant gastroschisis was diagnosed in the 32nd week of gestation. The entire bowel was floating in the amniotic fluid but the abdominal wall defect was small, liver and spleen were in the fetal abdomen and no other malformations could be visualized. This child was born by elective CS with immediate postnatal operation. In a review of 112 cases of omphalocele and gastroschisis (Kirk and Wah 1983), (118) no difference was found, between infants born vaginally or by CS, in mortality rate or time delay (in average 7 hours) from delivery to operation. In one vaginally delivered baby the bowel was damaged at birth. In this review with only 4 cases diagnosed prenatally the

mortality rate was 13,5% for gastroschisis and 29% for omphalocele. A major factor in mortality was concomitant occurrence of other anomalies seen most frequently in the omphalocele group. Others consider the mortality risk related mainly to the clinical condition of the baby at admission to the NICU (34, 125). In our opinion prenatal diagnosis and preparation for optimal surgical management at the delivery should lower the mortality rate. A prerequisite is referral of the mother to an obstetric department with pediatric surgery directly available. The route of delivery is still controversial but it seems reasonable to deliver cases with dense extra-corporeal organs by CS (26, 34, 76).

Part Four

Psychological reactions in women to prenatal diagnosis

In literature dealing with prenatal diagnosis of fetal malformations the reactions of the mother are seldom mentioned. It seems as if most research-workers have been engaged mainly in the diagnostic possibilities of ultrasound whereas maternal reactions and feelings have been disregarded. Psychological reactions in connection with perinatal loss, elective abortions or getting a malformed child are well documented (38, 46, 56, 104, 119, 126, 137, 146). Women's reactions to prenatal diagnosis have only been reported in connection with amniocentesis (19, 140, 194). Paper V and VI deal with the reactions of women who, after a routine ultrasound scanning, were informed that the fetus was malformed. These women were all unprepared and got a piece of information that they had never imagined.

The aim of routine ultrasound examinations is not primarily to discover fetal malformations. However, with more frequent application of routine ultrasound scanning and with advanced technique in ultrasound scanners, an increasing number of congenital abnormalities will be discovered already during pregnancy.

Prenatal diagnosis in the second trimester.

When the midwife suspects fetal malformation this must be confirmed by the obstetrician before informing the patient. More than one examination is often needed with a few days interval. Termination of pregnancy is performed as hysterotomy or intrauterine Rivanol instillation. A follow-up visit in the outpatient department is arranged one month later.

Mother-infant bonding may start already in the second trimester. A fetal loss at that time may cause a psychological trauma comparable to the reaction experienced after fetal/infant death near term (117, 172). The crisis reaction is quite similar in both situations (119).

The interview concentrated on the mothers' perception of given information, their decision to terminate the pregnancy and their psychological reactions. Most women felt that the medical information was correct in general. However, their capacity to assimilate traumatic information was limited. In the early phase of crisis the individual is confined and may often be unable to ask questions. Although most women were content with the initial information it is very important to give them an opportunity to react, discuss and ask questions a few days later.

Nine of 10 women did not find the decision to have a legal abortion too difficult. Information by the obstetrician that the child either would not survive or develop a severe handicap was important for their decision. However, in the coming years even smaller injuries or defects will be discovered in the 17th week of gestation. This will imply further demands on both the information and the support given by the obstetrician.

In our study seven women described conflicts between their principal attitude to legal abortions and their own experience in reality. Most women reported that a delay of a few days from a

decision-making to performing an abortion was necessary to get used to the fact that they should loose their baby.

Many women considered the routine post-abortion follow-up visit one month later at the obstetric department to be unsatisfactory because often another physician was then in charge. Some of the women reported the time of expected childbirth as especially difficult. In 1978, Cavenar et al described anniversary recurrent depressions and abdominal pain at time of expected childbirth in two women after a legal abortion (38). These problems were considered to be unconscious pregnancy fantasies from not working through the grief after the fetal loss. This is an important observation indicating that every woman should have an opportunity to meet her doctor if she some time after the abortion feels a need.

Most women experienced a severe psychological reaction when told the diagnosis. Those given warning signals (e.g. increased AFP in amniotic fluid, polyhydramnios or decreased fetal growth) reacted as strongly as those who had no prewarning. Similar reactions were observed by Lescot et al (1982) after diagnosis of fetal chromosomal anomalies by amniocentesis (140).

In the present study half of the women reported that they had not recovered completely at time of the interview, i.e. from 9 months to nearly 3 years after the abortion. They were still depressed and emotionally affected when talking about the abortion. One was primigravida and 5 women had more than one child previously. After the abortion four had given birth to healthy children, two were more than 17 weeks pregnant and two planned a new pregnancy. In five of these amniocentesis had been made. For three women the self-esteem was disturbed. Another three probably had similar feelings since they had achieved a higher self-esteem after the birth of a healthy child. This may reflect a need for the woman to undo, to prove her reproductive capacity and to work through feelings of guilt.

Prenatal diagnosis in the 32nd week of gestation.

Women, who after an ultrasound scanning in the 32nd week of gestation, are informed that their fetus is malformed, will be in quite a different situation i.e. without the possibility to have a legal abortion and thus "get rid of the problem". In the 32nd week the diagnosis leaves the woman with no alternative to the fact that she is going to give birth to a child with a very uncertain future.

As in early prenatal examinations a suspicion of fetal malformation by the midwife needs confirmation by the obstetrician who informs the woman. A definitive diagnosis often requires more than one examination. In the present investigation the woman was supported by regular follow-up until delivery. The delivery was planned and discussed with the parents actively involved in the decision-making process. There were no lethal malformations in this study and consequently no medical reason to discontinue the pregnancy. If postnatal survival is not possible the motivation to continue a pregnancy to term may be very small. Therefore it is of utmost importance that the parents are involved in the planning of the care.

The result in Paper VI showed that all 14 women preferred to get full information already during the pregnancy about the fetal malformation, possible treatment and prognosis. In the present study three were not informed before the delivery. They experienced this as a deceit and lost their confidence in the physician. These women were given correct medical information but could still not form a realistic picture of the baby. Some of them had the feeling that the baby could be "a kind of monster". This condition has been well described by D'arcy and Johns in mothers of malformed children, being separated immediately at delivery before they had had any possibility to see and touch their babies. The time delay until these mothers got information was described as the most distressing period with their imagination of the child's defect exceeding reality (48, 49). This is in accordance with the result

in the present study where most women found realities less frightening than fantasies.

However, feelings of ambivalence towards the child were expressed both during pregnancy and puerperium, i.e. wavering between the wish that the malformed child should die, and the vain hope for survival of a healthy baby. Naturally all women were unbalanced and depressed during the puerperium.

At time of the interview all women reported being in good mental balance. They were not emotionally upset when talking about the discover of the malformation and the child birth. There was no difference between women having a child with permanent sequelae (S-children) and those whose child was without sequelae (NS-children).

When asked if they would have considered an abortion at an earlier diagnosis, most women with S-children said that they would have chosen a termination and that they considered prenatal diagnosis important in a future pregnancy. However, none of the women with NS-children considered that the malformation of their child should justify an abortion.

After delivery of the malformed child, three had given birth to healthy children and one was pregnant, 3 mothers with S-children and one with a NS-child previously.

For a few parents a deteriorated relationship had developed but for most it was unchanged or even improved. At the time of the interview the women did not express feelings of guilt and inferiority. It seems as if these women have solved both practical and emotional problems strengthening their self-esteem. Still, psychological self-defence may of cause have repressed some of the more negative feelings.

General Discussion

The aim of the present investigation was neither to defend nor to reject routine screening by ultrasound in every pregnancy. This thesis may shed some light on positive and negative effects of the prenatal diagnosis achieved in a screening population.

However, it is necessary to consider the possible side effects of ultrasound. In 1982, WHO published a report summarizing the results of experimental studies in cell cultures, experimental animals and human fetuses (91). A linear scanner gives an intensity of $0,06-10 \text{ mW/cm}^2$ (SATA). In 1981, Liebeskind et al, by using 15 mW/cm^2 and a total exposure time of 30 min, could demonstrate changes in the ultrastructure of fibroblasts in rats (147). Suppression of cell growth of human cells in amniotic fluid was detected by Maeda and Murao 1977 with 60 mW/cm^2 and 30 min exposure time (156). In 1977 and 1980, Hara et al found an increased incidence of fetal malformations in rat offspring after 2000 mW/cm^2 and 5 min exposure time (86). Moore et al suggested in 1982 that reduced birth weight could be found among children who had been exposed to ultrasound (162). Recently, Stark et al published the results of a multivariate analysis for perinatal, physical and developmental factors to a mean age of 9 years in groups of children exposed or not exposed to prenatal ultrasound. They found no significant differences between the two groups although the exposed group had more pregnancy complications which in most cases had been the indication of the ultrasound examinations (196). In most experimental studies both intensity and exposure time have differed from that used in diagnostic ultrasound. No certain harmful effects have been reported in human beings in spite of the frequent use of ultrasound.

To make an accurate prenatal diagnosis it is peremptory to have good knowledge of normal anatomy to enable a distinction between normal and pathological. In Paper I standard growth parameters were

obtained for brain structures of great importance for diagnosis of abnormalities. The results showed that it is possible to make such measurements routinely in 90% of the cases. The growth of LVW and the LVW/HW are in good accordance with previous reports with LVW/HW-index declining from 46% in gestational week 17th to 28% at term. The results showed that the growth of LVW was correlated to gestational week more than to birth weight. The difference between boys and girls might be a reflection of the biological variation normally found post term (212). However, the differences found were less than one standard deviation which implies that mean values for the total population can be used for both sexes.

The normal material in Paper I proved valuable for the diagnosis of brain ventricle dilatation. The 0.12% incidence of ventricle dilatation during the period 1980-84 is comparable to an estimated of 0,14% (134). It is reasonable to presume that the intrauterine incidence is lower since cases of postnatally acquired ventricle dilatation may develop (see Paper III).

The development of the ventricle dilatation during pregnancy and the later outcome for the children have been registered in all cases. Only through knowledge of longterm outcome is it possible to improve the information given to parents both in case of early or late diagnosis. Intrauterine treatment of hydrocephalus has opened new therapeutic possibilities. However, in Sweden probably only a few cases will be suitable for such treatment. Because of a widespread use of routine ultrasound scanning early prenatal diagnosis will become more frequent. In a case of pronounced hydrocephalus, however, today only few parents will prefer to continue the pregnancy. In a case of late diagnosis, improved neonatal care will imply that preterm delivery is preferable if progressive hydrocephalus is developing. Not long ago cephalocentesis was the method of choice in such cases still reported in the literature (132). In the 1970's reports correlating the thickness of the brain parenchyma to later outcome recommended cephalocentesis if it was less

than 5-10 mm (95, 132). However, in a study from 1974 of shunt-operated hydrocephalic children, Raimondi and Soare found the IQ unrelated to the thickness of the cerebral mantle (177). We therefore consider it important to evaluate time and method of delivery in the individual case in close collaboration with the pediatrician and neurosurgeon.

Paper II and IV demonstrate some prenatal malformations which can be discovered by ultrasound. These studies comprise the total population of pregnant women thereby achieving incidences of these abnormalities. The attendance of pregnant women during these years was almost total. No postnatal reports about false negative ultrasound examination have appeared in this population neither for ventricle dilatation, nor intestinal or urinary tract malformations.

In most cases of intestinal and urinary tract malformations the situation is different since several of these malformations can be treated by surgery and the prognosis for the child is favorable in many cases. The result of surgical intervention will often depend on the state of the child at time of admission. Thus, prenatal diagnosis implies a possibility for an improved preoperative planning being especially important for intestinal tract malformations.

The prognosis for a unilateral urinary tract lesion is usually good. Still, a prenatal diagnosis and early postnatal treatment will be beneficial especially in cases of obstructive nephropathies. In bilateral obstruction the renal function may be estimated from the degree of bladder distension, the amount of amniotic fluid and the renal parenchyma thickness (84, 88, 138). These measurements may be valuable in deciding the optimal time of delivery. In very preterm infants intrauterine treatment may be indicated. Still, many uncertainties exist, e.g. it is known that during the pregnancy transient hydronephrosis later resolving may be seen (84, 88). In these cases repeated ultrasound scanings increase the possibility to discern normal from abnormal or transient from persistent aberrations.

The study in Paper III demonstrates a further application of diagnostic ultrasound in perinatal medicine. The study group of infants developing brain ventricle dilatation during the neonatal period was selected by application of the normal growth curves presented in Paper I. Twenty of 27 infants in this group developed ventricle dilatation, probably a complication of an intracranial hemorrhage which is generally regarded as the most common cause in the postnatal period (2, 97, 143, 187, 206, 209). Analysis of perinatal factors in preterm and term infants with ventricle dilatation showed a significant correlation between poor outcome and intrauterine and neonatal asphyxia ($p < 0,01$). This is also in accordance with prevailing theories about the etiology of intracranial hemorrhages (206, 209). Brain ventricle dilatation implied a less favorable outcome in this study of severely ill newborns. Knowledge of the outcome in these severely ill infants is important not only for the pediatrician but also for the obstetrician in the clinical evaluation of pregnancy and delivery complications.

In our experience, ultrasound screening of every pregnancy will imply a 45% discovery rate of serious malformations occurring in about 1% of pregnancies. Some of these cases should probably be diagnosed without routine screening since malformation often influences fetal growth and a concomitant oligo- or polyhydramnios will imply an abnormal decreased or increased size of the uterus (39, 40, 132, 171). Furthermore, ultrasound examinations for other pregnancy complications may disclose additional abnormalities. Thus, while accepting ultrasound as a valuable diagnostic tool we must also be ready for unexpected disclosure of fetal malformations.

The results of our retrospective investigations (Paper V and VI) of psychological reactions to prenatal diagnosis of fetal malformation showed that the women prefer to be informed immediately in connection with disclosure of the abnormality. They want an open and honest discussion. When the diagnosis was made in the third trimester the remaining part of the pregnancy was experienced as almost

unbearable. Still, the women considered that a delay of information until delivery would have been even more terrible.

Early diagnosis of severe malformations and termination of the pregnancy is generally regarded as a positive effect of prenatal diagnosis (20, 39, 40, 95, 98, 132). The findings in Paper V indicate that although a practical problem can be solved this solution leaves deep scars in the mind of the woman. Half of the women had still not recovered several months or years after this traumatic experience. These women need compassion and emotional support from the obstetrician and from their relatives. In contrast the women in the second group with late prenatal diagnosis seemed less mentally unbalanced at time of the interview. Perhaps they have had more opportunity than women with selective abortions to work through their experience. The use of ultrasound is not only a question of developing a satisfactory technique; the psychological consequences must be considered as well.

General Conclusion

The findings of this thesis have shown the following.

- Growth curves for normal brain lateral ventricular size and development during the 2nd and 3rd trimester of pregnancy in a consecutive population.
- Prenatal diagnosis of brain ventricular dilatation consequently can be made with certainty at mid-gestation and in late pregnancy.
- Brain ventricle dilatation in ill newborn infants was mainly related to perinatal asphyxia and postnatal complications.
- Prenatal ultrasound diagnoses of intestinal and urinary tract malformations imply improved management after delivery.
- Prenatal diagnosis or suspicion of abnormality is a heavy burden on the woman and her family during pregnancy and many years after termination of pregnancy or delivery. The situation is best handled in an atmosphere wherein feelings of emergency or urgency are minimized and time for discussion maximized.
- Routine ultrasound scanning is not only a question of technical skill and capacity. Knowledge about fetal growth and longterm outcome for the child is necessary as well. Besides counselling, insight, compassion and emotional support from the obstetrician and midwife are equally important for optimal management. This also requires close co-operation between obstetrician, neonatologist, pediatric-surgeon and neuro-surgeon.

Appendix

Table I. A survey of pre- and postnatal ultrasound examinations in this investigation. US = ultrasound, m = months, y = years

Paper	I	II	(II)	III	IV	V and VI
US-examinations:						
Year	1983	1980-81	1980-84	1980-83	1980-82	1980-83
Length of investigation	4m	13m	4y	32m	24m	34m
Population no.	654	3120	11146	166	6020	8422
Fetal/neonatal abnormalities	-	5	13	27	6	27

Table II. Obstetrical and neonatal data of the 27 infants with brain ventricle dilatation neonatally. The ultrasound diagnosis correlated to computer tomography, analysis of cerebrospinal fluid achieved at lumbar puncture and postmortem examination.

Case no.	Pregnancy complications	CTG + or - ominous	Mode of delivery	Gestational age at delivery (weeks)	Birth weight g	Sex	Apgar score	EEG normal or pathological	Seizures + or -	Pulmonary complications
1	Hemorrhages		V	25	750	f	2-7-8	pathological	-	BPD
2	Twins	-	S	27	970	f	3-4-6	normal	-	BPD, pneumoth
3	Abruptio pl.		S	26	800	m	2-5-7	pathological	+	BPD, pneumoth
4	-		S	27	850	m	7-8-8	pathological	+	HMD, pneumoth
5	Abruptio pl.	+	S	28	700	m	5-7-8	-	-	BPD
6	Abruptio pl.		S	28	1300	m	5-7-7	pathological	+	BPD, pneumoth
7	Preeclampsia	-	S	29	940	m	2-9-	normal	-	BPD
8	Abruptio pl.	+	S	29	970	m	9-	-	-	BPD, pneumoth
9	IUGR	+	S	30	720	f	1-6-8	-	+	BPD
10	Abruptio pl.	+	S	30	1400	m	6-8-9	-	-	Pneumoth
11	Abruptio pl.		S	31	2185	m	7-9-9	patological	+	IRDS, pneumoth
12	Twins		S	31	1470	m	9-10-	normal	-	PMA, pneumonia
13	Preeclampsia	+	S	31	975	m	9-9-9	pathological	+	Apnea
14	Preeclampsia	+	S	32	830	f	1-7-	pathological	+	BPD
15	Severe Rh-imm	+	S	32	1980	f	3-5-6	pathological	+	
16	-	-	V	33	2000	m	3-5-6	pathological	+	Pneumonia
17	Preeclampsia	+	S	34	1840	m	10-10-	patholog-normal	+	HMD, pneumoth
18	Hemorrhage	-	S	34	2420	f	9-10-10	pathological	+	IRDS
19	Abruptio pl.	+	S	36	2010	f	0-5-6	pathological improved	+	PMA
20	Preeclampsia	+	S	36	2170	m	1-6-7	pathological	+	PMA
21	-	-	V	38	3390	m	4-7-7	normal	-	-
22	Hydramnios	-	V	38	3000	f	3-	pathological	+	-
23	Preeclampsia	+	S	38	2060	f	3-6-7	pathological	+	-
24	-	-	V	40	3374	f	9-10-10	normal	-	-
25	-	+	S	41	3500	m	2-4-4	pathological-imp	-	-
26	IUGR susp	+	S	40	2490	m	0-3-7	pathological improved	+	-
27	-	-	V	42	3210	f	9-10-10	pathological	+	-

Abruptio pl. = abruptio placentae Rh-imm = Rh-immunization

IUGR = intrauterine growth retardation

S = Cesarean section V = vaginal delivery f = female m = male

BPD = bronchopulmonary dysplasia pneumoth = pneumothorax

HMD = hyaline membrane disease

IRDS = idiopathic respiratory distress syndrome

PMA = pulmonary maladaptation

Assisted ventilat. days	Ultrasound day after delivery and diagnosis	Computer tomography day after delivery and diagnosis	LP with CSF analysis	Postm.examination day after delivery and diagnosis	Outcome
43	28 BVD	80 BVD	TP	-	Moderate CP
30	4 ICH 13 BVD	-	-	-	Healthy
17	10 ICH, BVD	-	-	18 ICH, BVD	-
35	1 ICH 6 BVD	75 BVD susp. ICH	sidero-phages	-	Moderate CP
5	109 BVD, atrophy	-	-	-	Severe CP
58	2 ICH, BVD	40 ICH, BVD	sidero-phages	65 ICH, BVD	-
60	46 ICH, BVD	87 ICH, BVD	-	214 No postmortem examination	-
113	3 ICH 15 BVD	44 BVD	-	109 ICH, BVD	-
56	81 BVD, atrophy	-	-	140 BVD, atrophy	-
8	10 ICH, BVD	-	sidero-phages	-	Light CP
19	4 ICH, BVD	-	TP	-	Moderate CP
4	34 ICH, BVD	52 BVD	sidero-phages	-	Moderate CP
0	24 ICH, BVD	168 BVD	-	-	Severe CP
26	11 BVD	-	TP	28 ICH, BVD	-
20	7 ICH, BVD	-	-	34 ICH	-
33	3 ICH 19 BVD	45 BVD	sidero-phages	52 ICH, BVD Corpus callosum agenesis	-
5	20 ICH, BVD	30 BVD	sidero-phages	-	Light CP
12	4 ICH 6 BVD	-	-	12 ICH, BVD	-
31	5 ICH, atrophy	28 BVD, atrophy	TP	31 ICH, atrophy Leuconalaci	-
11	4 ICH 15 BVD	39 BVD	-	330 BVD	-
35	15 BVD	4 BVD	TP	-	Light CP
44	7 BVD 34 Atrophy	-	TP	43 Subdural hemorrh. Atrophy, Dystrophia Myotonica	-
19	6 ICH 19 BVD	135 BVD	sidero-phages	-	Severe CP
0	16 BVD	12 BVD	-	-	Moderate CP
10	2 ICH, atrophy	-	-	127 Atrophy	-
1	2 ICH 6 BVD	18 BVD	TP	-	Moderate CP
3	25 ICH, BVD	19 ICH, BVD	-	-	Severe CP

LP = lumbal puncture with analysis of cerebrospinal fluid (CSF)

BVD = brain ventricle dilatation

ICH = intracranial hemorrhage

TP = elevated total protein in CSF

CP = cerebral palsy

Questionnaire on fetal malformation discovered in the second trimester.

Name, age, education

Previous pregnancies: Number of children

Legal and spontaneous abortions

Ectopic pregnancies

Treatment for infertility

Other information of interest

- 1) Did you experience any reaction on the part of the midwife?
Did you suspect that something was wrong?
- 2) How did you react being told that the fetus was malformed?
- 3) Did you get sufficient information from the doctor about the malformation? Did you have a need for asking questions?
- 4) You were informed of the fetal malformation directly after the ultrasound examination. Would you have preferred to get the information gradually in connection with further visits at the department?
- 5) Were you accompanied by your husband when you received the information?

if no: Had it been better to postpone the information until he could have joined you?
- 6) Did you ever take into consideration to continue the pregnancy?
- 7) Did you find it difficult to decide to terminate the pregnancy?
- 8) To whom did you confide yourself during this period? Did you get sufficient support?

9) What was previously your attitude to legal abortion, especially in the second trimester?

10) Would you ever consider late abortion again?

11) The time between your decision to have an abortion and the operation, was it too long?

Do you suggest any improvement?

12) How long did it take to work through the abortion?
Have you recovered?

13) Could anything have been of help to you?

14) With whom was it possible for you or/and did you want to talk after the abortion?

15) How did you experience the visit at the department 1 month after the abortion?

Would more contact with doctors or socialworkers have been valuable?

16) Did you see the aborted fetus?

if no: Do you regret that you did not?

if yes: What were your feelings looking at it?

17) Have you ever considered that you did not make the right decision by an abortion?

18) Have you been pregnant after the abortion?

if yes: What was the outcome? Were you more worried and anxious during this pregnancy?

Did you have an ultrasound examination in the 17th week of gestation?

Did this examination make you feel more reassured for the rest of the pregnancy? Did you have an amniocentesis?

if no: Why have you not attempted a new pregnancy?

19) Are you planning to have children in the future?

if yes: Will you ask for an amniocentesis?

if no: Why will you not attempt a new pregnancy?

20) Having been pregnant with a malformed fetus has this in any way influenced your self-esteem?

if yes: How?

21) Has it caused any change in your relations with your husband?

Has it influenced your sexual life?

Questionnaire on fetal malformation discovered in the 32nd week of gestation.

Name, age, education

Previous pregnancies: Number of children

Legal and spontaneous abortion

Ectopic pregnancies

Treatment for infertility

Other information of interest

- 1) How did the information that your baby was injured influence the rest of the pregnancy?
- 2) Was the information sufficient, too detailed or too superficial?
- 3) Did you get a too positive or too negative picture of your baby?
- 4) Did you find it difficult to understand the consequences of the information?

Did you experience the baby as "a kind of monster"?

- 5) Would you have preferred not to be informed during the pregnancy?
- 6) Did you get sufficient support during the pregnancy?
From whom?

What did you miss?

- 7) Method of delivery CS: Would you have preferred a vaginal delivery?
V: Would you have preferred a CS?

How did you experience the delivery? CS: V:

Analgesic during the delivery and during the puerperium

- 8) Did you feel mentally unbalanced during the puerperium?
- 9) How long did it take before you felt mentally balanced?

10) How do you feel now?

11) How is the day-to-day life with your child?

Have you experienced problems in the relationship to your
other children?

parents?

fellow workers?

Have you had problems with the child care?

Has it influenced your own education?

12) Have you got sufficient support from the hospital?

What was lacking?

13) Do you now regard your child as handicapped?

14) Do you have any notion of your child's future?

15) If you had had the information of your child's injury already
after the ultrasound examination in the 17th week of gesta-
tion, would you have considered an abortion?

16) Has it caused any change in your relationship to your husband?
Has it influenced your sexual life?

17) Have you been pregnant again or do you plan to have children
in the future?

18) Will you ask for an amniocentesis in case of a future
pregnancy?

Did you have amniocentesis in your previous pregnancy?

19) For women having a subsequent pregnancy

Did you feel very anxious during the later pregnancy?

20) Did the ultrasound examination in the 17th and 32nd week of
gestation make you feel more reassured during the rest of the
pregnancy?

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I

Ultrasound measurement of the fetal cerebral ventricles ~ A prospective, consecutive study

C. Jörgensen, I. Ingemarsson, E. Svalenius,
S. Montan & N.W. Svenningsen

Summary

In a consecutive study of 654 pregnant women real-time ultrasound was used for measurements of the fetal biparietal diameter(BPD), abdominal diameter(AD), femur length(FL), cerebral lateral ventricle width(LVW) and cerebral hemisphere width(HW) at gestational weeks 17 and 32. In 408 cases an additional examination was made in week 39 and in postterm pregnancies re-examination was made once a week until delivery. The LVW and HW growths were correlated to gestational week, BPD and birthweight.

The LVW and HW increase as linear regression lines relative to biparietal diameter. From these regression lines the curve for the ventricle/hemisphere (LVW/HW) index % was calculated.

From 17 to 32 weeks of gestation both LVW and HW increase and LVW/HW-indices decrease significantly. From 32 weeks to term, the change is less for all parameters and mainly related to HW growth, i.e. increased width of brain parenchyma estimated from LVW minus HW.

In preterm and term infants with birth weight under 2800g comparisons of fetal LVW and HW in equal gestational weeks showed no differences. Similar comparisons were made between small and large term infants and no difference was found at gestational week 32 and week 39, but at week 39 only for birth weight ranging from 3000-4500g.

In fetuses deviating more than $\pm 2SD$ in these measurements weekly re-examinations are necessary for exact diagnosis of abnormalities.

Introduction

Abnormalities in the growth of the fetal brain structures may have wide implications on the outcome of the pregnancy. Because of compressibility of the young fetal hydrocephalic head ultrasound

measurement of the fetal biparietal diameter (BPD) is inadequate for diagnosis of a prenatal hydrocephalus. The BPD is often within normal range and may not increase significantly until after 30 weeks of gestation (1).

Previous studies of the fetal brain ventricles have presented ranges for various parameters at different gestational ages (2, 3,4,5,6). A longitudinal study of the postnatal ventricle growth has been made by Levene (7) in preterm infants.

The aim of our present investigation was to study the growth of the fetal brain ventricles and hemispheres longitudinally in gestational week 17, 32 and from 39 until delivery and to obtain standard intrauterine fetal brain growth parameters.

Material and Methods

In the referral area of the University of Lund every pregnant women is offered two routine ultrasound examinations at gestational week 17 and at week 32. The total population of 654 pregnant women with expected delivery date between January 10th to May 10th 1983 were examined routinely in the 17th and 32nd week of gestation. To achieve growth parameters from the last part of the pregnancy an additional examination was made in gestational week 39 (38-40 weeks) in 408 (62,4%) of the women, 146 women were delivered before week 39 and 100 women refused further examinations. A number of 51 postterm pregnancies were followed once a week till delivery. No fetal malformations were included in the study, whereas patients suffering from diabetes mellitus, hypertension, as well as those who later developed preeclampsia, placenta insufficiency or other pregnancy complications, entered the study in a number corresponding to the incidence in our pregnant population. The ultrasound examinations were performed by specially trained midwives using a real-time ultrasound scanner (Hitachi EBU 22 or 25).

The following parameters were measured: Biparietal diameter (BPD), abdominal diameter (AD) and femur length (FL). The gestational age of the fetus was decided based on normal range curves for BPD (8).

The hemisphere width (HW) and the width of the anterior horns of the lateral ventricles (LVW) were measured at the level where the lateral wall of the anterior horns of the lateral ventricles appear as two short lines parallel to the midline echo, i.e. LVW from the midline to the first strong echo of the lateral wall and at the same level HW from the middle of the midline echo to the first strong echo of the inner table of the skull as illustrated in Fig.1.

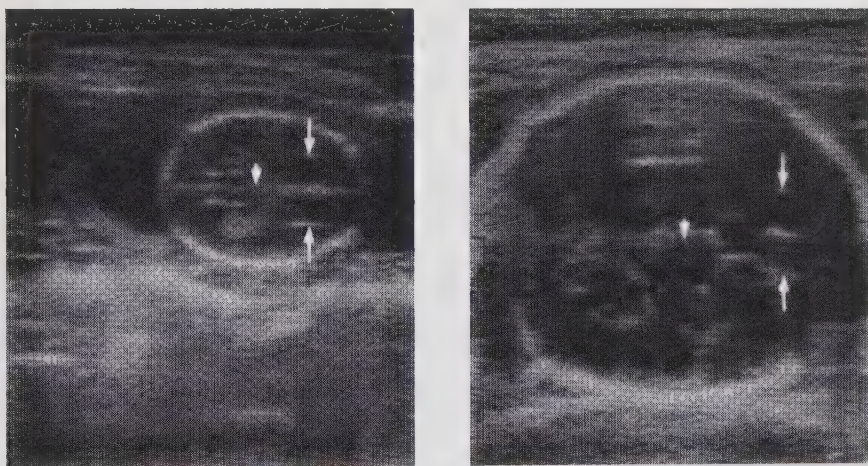


Fig. 1. Frontal horns (arrows) and midecho (arrow head) in transverse section. Gestational week 17 (a) and 32 (b).

Both parameters, indicated in the results for one hemisphere, are based on measurement on both sides and calculation of the mean value. The ventricle/hemisphere index (LVW/HW) was calculated from these parameters. All parameters were measured in gestational week 17, 32, 39 and once a week until delivery.

LVW could not be visualized and adequately measured when BPD was measured in 13,6% at week 17, 9,1% at week 32 and 13,7% at week 39. At least one measurement was made in every fetus. In a few cases BPD could not be measured adequately at the previously mentioned

gestational weeks and in other cases the women came for the examination at the wrong gestational week, explaining the difference between total number of patients (654) and the number given in Table I.

Statistical methods: The data were computer analyzed resulting in mean, standard deviation (SD) and standard error of the mean (SEM) for LVW, HW and LVW/HW according to every represented BPD (BPD 26mm to 50mm and 74mm to 103mm), gestational weeks (17, 32, 33, 38, 39, 40, 41 and 42) and birth weight at delivery. Student T-test was used when comparing LVW, HW and BPD for boys and girls, term to preterm infants with birth weight in both groups under 2800g, and term infants within different birth weight groups.

Results

Fig.2 and Table 1 show the meanvalue, the SD and the SEM for LVW, HW and LVW/HW-index in different gestational weeks. The mean LVW increases little from week 32 (11,0mm) to week 39 (12,0mm) and to week 42 (12,5mm) whereas the mean HW increases from week 32 (39,0mm) to week 39 (45,2mm) and to week 42 (47,3mm). Although HW increases slightly the LVW/HW-index is almost unchanged from week 38 to 42. From a value of 46% at week 17 the LVW/HW-index decreases to 28% at week 32 and to 27% at term.

The mean and standard deviation of LVW, HW and LVW/HW were calculated for every corresponding BPD (i.e. for BPD 26mm to 50mm and for 74mm to 103mm) as shown in Fig.3. LVW and HW were correlated to BPD with linear regression characterized as follows: $LVW = b(LVW) + mX(LVW)$ and $HW = b(HW) + mX(HW)$, where b = intersection with the Y-axis and m = slope for the line. For these lines we found a correlation coefficient for LVW: $r = 0,966$ and for HW: $r = 0,998$. The coefficient of this program is defined ranging from -1 to +1, indicating a high positive correlation when r is near +1. The values 0,966 and 0,998 therefore represent a very high positive correlation.

Table I. Mean, standard deviation (SD) and standard error of the mean (SEM) for LVW (mm), HW (mm) and LVW/HW-index (%) in relation to weeks of gestation.

Gestation week	No. of fetuses	LVW mm			HW mm			LVW/HW-index %		
		Mean	SD	SEM	Mean	SD	SEM	Mean	SD	SEM
17	557	7,8	1,0	0,0	17,1	2,2	0,1	45,9	6,4	0,3
32	515	11,0	1,0	0,0	39,0	2,1	0,1	28,3	2,7	0,1
33	61	11,3	1,0	0,1	40,5	2,0	0,3	28,2	2,5	0,3
38	59	11,9	1,5	0,2	44,8	2,2	0,3	26,4	3,1	0,4
39	263	12,0	1,7	0,1	45,2	2,2	0,1	26,6	3,7	0,2
40	24	12,9	1,4	0,3	46,5	1,6	0,3	27,8	3,3	0,6
41	16	12,1	1,3	0,3	46,1	1,9	0,5	26,3	3,1	0,7
42	19	12,5	1,7	0,4	47,3	2,3	0,5	26,5	3,4	0,7

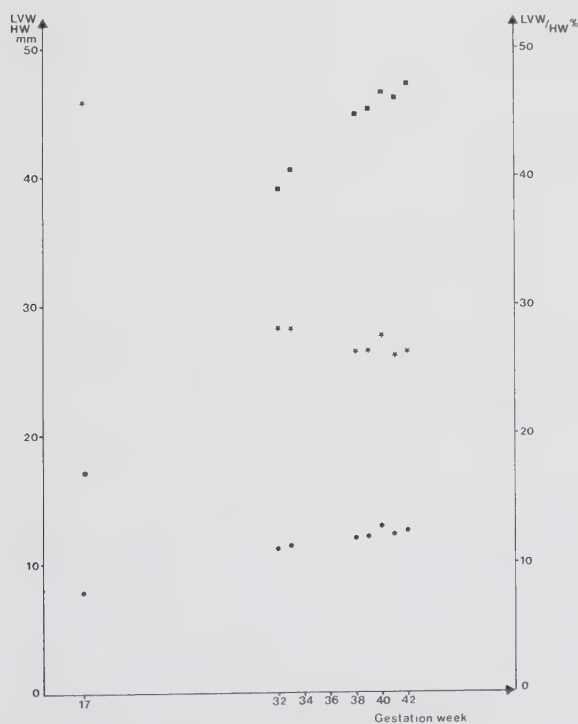


Fig. 2. Mean values of LVW (mm) ● , HW (mm) ■ and LVW/HW-index (%) ★ against week of gestation.

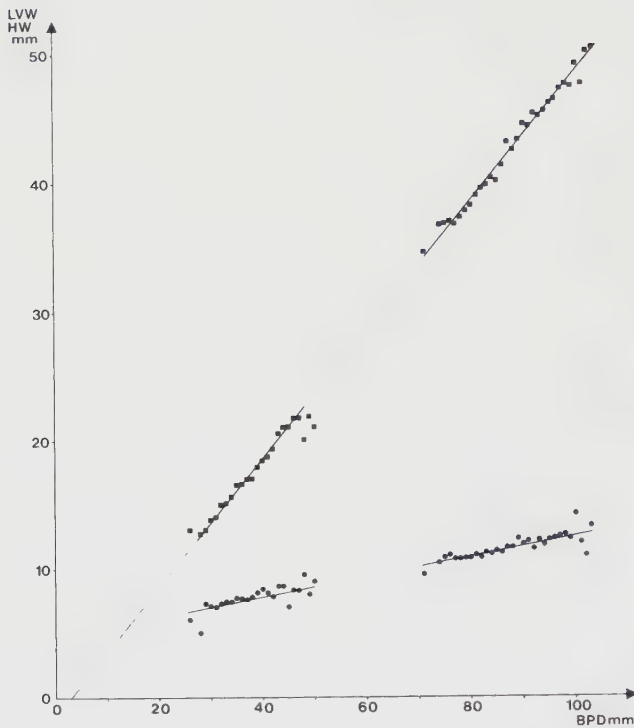


Fig. 3. Linear regression lines for LVW (mm) ● and HW (mm) ■ in relation to the corresponding BPD (mm).

As the patients examined at the beginning of the 2nd trimester (period s = BPD 26mm to 50mm) and in the 3rd trimester of the pregnancy (period e = BPD 74mm to 103mm) do not represent the same sample of patients, the values for LVW and HW were calculated against BPD for the two periods individually.

LVW:	BPD 26mm to 50mm	bs = 3,73	ms = 0,10	rs = 0,79
	BPD 74mm to 103mm	be = 4,21	me = 0,08	re = 0,83
	BPD 26mm to 103mm	b = 4,63	m = 0,079	r = 0,966
HW:	BPD 26mm to 50mm	bs = 1,39	ms = 0,42	rs = 0,98
	BPD 74mm to 103mm	be = -0,62	me = 0,49	re = 0,99
	BPD 26mm to 103mm	b = -1,53	m = 0,499	r = 0,998

The values are so close to each other that we have accepted all observations to be representative for the main population. The number of patients in the present study is 654 compared with 112 to 200 patients in other studies (2, 3, 4, 6).

From values of LVW and HW the corresponding LVW/HW-index was calculated and the curve constructed from the equations $LVW = 4,63 + 0,079 \times BPD$ and $HW = -1,53 + 0,499 \times BPD$ as follows:

$$\frac{LVW}{HW} = \frac{4,63 + 0,079 \times BPD}{-1,53 + 0,499 \times BPD} = \frac{0,079(58,6 + BPD)}{0,499(-3,07 + BPD)} = \frac{(BPD + 58,6)}{(BPD - 3,07)} = 15,83 \dots \%$$

Fig.4. Even this construction is encumbered with a small error by not using the four individual regression lines, but the error will not be more than maximal 3,5% or well below the standard deviation.

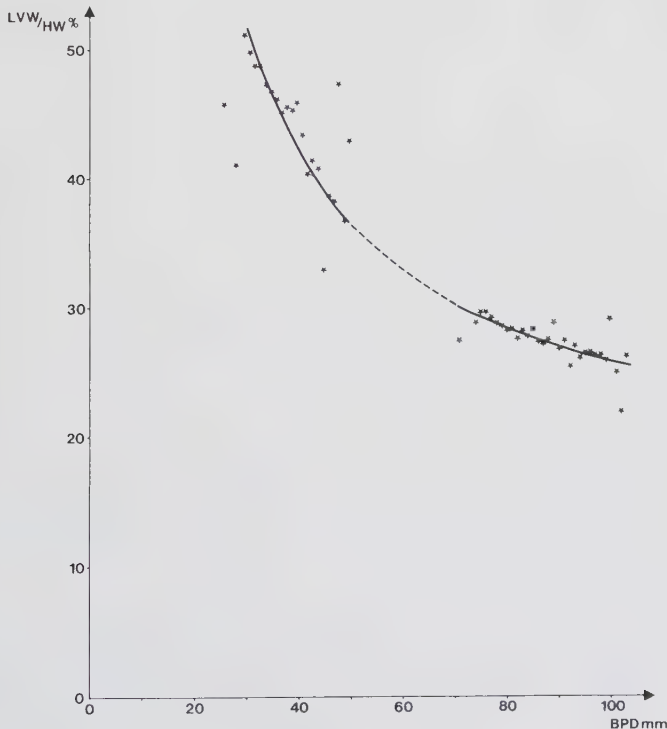


Fig. 4. LVW/HW-index (%)★ calculated for the corresponding BPD (mm).

Table II shows LVW, HW and LVW/HW-index in week 32 and 39 related to birth weight at delivery. The LVW did not correlate to birth weight neither in gestational week 32 nor at week 39 for birth weight ranging from 2501g to 4500g. In contrast there was an increased HW with increasing birth weight both in week 32 and 39.

Table II. LVW (mm), HW (mm) and LVW/HW-index (%) at gestational week 32 and 39 in relation to birth weight.

Birth weight g	LVW mm		HW mm		LVW/HW %		No. fetuses	
	week of gestation		week of gestation		week of gestation		week of gestation	
	32	39	32	39	32	39	32	39
< 2500	11,6	13,0	37,4	43,0	31,0	30,0	7	1
2501-3000	10,8	11,2	37,5	43,8	28,7	25,2	32	16
3001-3500	10,9	12,1	38,8	44,5	28,2	27,1	139	79
3501-4000	11,0	11,9	39,1	45,5	28,2	26,1	175	103
4001-4500	11,0	12,0	39,6	45,7	28,0	26,2	86	52
> 4500	11,8	13,4	41,0	48,2	29,0	27,8	19	12

Comparing the values for boys and girls separately it was found (Student's T for non-paired observations) that boys had larger LVW than girls in week 17 (mean = 7,9mm versus mean = 7,7mm; $p < 0,01$) and week 32 (mean = 11,1mm versus mean = 10,9mm; $p < 0,01$). Boys also had larger HW than girls but significant only in week 32 (mean = 39,3mm versus mean = 38,8mm; $p < 0,005$). Even BPD values were larger for boys than girls both in week 17 (mean = 37,5mm versus mean 36,4mm; $p < 0,01$) and week 32 (mean = 82,8mm versus mean = 80,9mm; $p < 0,01$). The same tendency was seen in all the other examination weeks but no significant difference was found probably caused by a much smaller number of observations.

The 13 fetuses with LVW, HW and LVW/HW indices deviating more than $\pm 2SD$ from the mean have been evaluated separately regarding their growth pattern. No specific pattern could be found. In all these fetuses only a single LVW or HW measurement was deviating

more than $\pm 2SD$ from the meanvalue, whereas previous or following measurements were normal. At birth all except 1 were within $\pm 2SD$ at birth. The infant, having LVW above $\pm 2SD$ in week 32 and 39, showed no signs of hydrocephalus at delivery or at follow-up examinations up to 8 months of age.

The growth of LVW in 26 intrauterine growth retarded term infants (i.e. 40 weeks of gestation ± 14 day) was compared with the growth in 24 preterm infants (i.e. born before term - 28 days). Birth weights in both groups were less than 2800g. At equal gestational weeks the same LVW values were found in both groups.

Discussion

The cerebral ventricles of the fetus can be visualized distinctly by high-resolution real-time ultrasound scanners (9, 10, 11, 12, 13). This is possible even in a routine scanning program performed by trained midwives (14). Thereby a prenatal diagnosis of fetal brain ventricle dilatation is possible (15). As shown in this study ultrasonic measurements of LVW and HW can be included in a routine screening program. These structures (LVW and HW) could be visualized and adequately measured in 86,3% to 90,9% of all patients depending on gestational week. This rate of successful measurements is slightly better than previously reported (3). The reason for failure is usually the fetal position, particularly when the sagittal suture is in the anterior-posterior plan. In some cases a measurement is still possible by external gentle manipulation.

Detection of fetal cerebral ventricle dilatation in the first half of the pregnancy makes a legal termination possible. In a previous study (14) the incidence of fetal brain ventricle dilatation was 0,16%. In a population of 3120 pregnant women 5 hydrocephalic fetuses were found and early diagnosis (and termination) was made in one of these.

The aim of this prospective study was to achieve standard growth parameters of the fetal brain growth. The lateral ventricle width (LVW) is larger in week 17 than later in the pregnancy with LVW/HW-index declining from 46% in week 17 to 27% at term. These findings confirm other studies with similar observations (2, 3, 4, 6). From week 32 to term the LVW increase is small in contrast to the HW increase which follows the BPD curve.

In a comparison between preterm infants and term infants with birth weight in both groups < 2800g it was shown that LVW at 17 and 32 weeks of gestation was not changed in the latter group of term infants with retarded fetal growth.

Even comparison of birth weight groups of term infants showed that LVW to a great extent was independent of birth weight. Thus, our results should hardly be influenced by using consecutive pregnancies (including patients with complicated pregnancies where the fetal growth might be affected) rather than selective normal pregnancies. It may even be concluded that LVW is correlated to gestational week rather than birth weight.

Denkhaus and Winsberg (2) have reported normal values for measurement of the bifrontal horn widths. Regarding their curve for the relation between bifrontal width and BPD it can be seen that it is concordant with a linear regression. Our results are very similar, differing less than 1mm when adapted to bifrontal horns measurement, for every represented BPD. Therefore it seems reasonable to assume that the regression lines in Fig.3 and 4 are representative even for the intermediate BPD area (51mm to 70mm).

Johnson et al (3) Hadlock et al (4) and Jeanty et al (6) have reported normal values for measurement of the midbody of the lateral ventricle. Hadlock et al compared their results for LVW/HW midbody to Johnson et al using the same technique. For the period 27-40 weeks of gestation they have mean 0,30, Johnson et al 0,29

and in this study 0,27 using LVW/HW for BPD 71-103mm covering the same gestational period. The difference 0,03 might be caused by the different method of measurement in our study.

The boy vs. girl differences of BPD and corresponding HW and LVW were significant for gestational weeks with a high number of observations. With a low number of observations a significant difference could not be found although a similar tendency existed for all gestational weeks. It is well known that boys have slightly larger head circumference curves after birth (16). We think that the results in the present study may reflect the phenomenon that biological variation remains substantially unchanged before, at and after term (16).

Some infants had occasionally a single fetal brain parameter more than $\pm$ 1SD and $\pm$ 2SD. In all but one the postnatal follow-up examination showed values within the normal range. Fluctuations of ventricle size might be due to incorrect measurement. However, alterations in fetal cerebral blood flow as well as cranial compression should also be taken into consideration (7).

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II

Prenatal diagnosis of fetal brain ventricle dilatation

C. Jørgensen, I. Ingemarsson & N.W. Svenningsen



Prenatal diagnosis of fetal brain ventricle dilatation

CONNIE JÖRGENSEN, INGEMAR INGEMARSSON & NILS W. SVENNINGSSEN

Departments of Obstetrics and Gynecology and Paediatrics, University Hospital, Lund, Sweden

Summary. In a 13 month prospective screening programme 3120 pregnant women were examined with diagnostic ultrasound at 17 and 32 weeks gestation; five fetuses (0.16%) were found with dilatation of brain ventricles. Early diagnosis was made in one patient resulting in legal termination of the pregnancy. The four other patients were diagnosed at the second ultrasound examination.

Modern ultrasound technique has made it possible to investigate intracranial structures of the fetus and fetal intracranial anatomy has been studied in detail (Kossoff *et al.* 1974; Johnson *et al.* (1975). Garrett *et al.* (1975) described the characteristic ultrasound picture of fetal hydrocephalus. Various fetal intracranial abnormalities have now been diagnosed by ultrasound (Hobbins *et al.* 1979; Little & Campbell 1980). However, these reports originate from referral centres and the true incidence of fetal brain ventricular dilatation in a normal pregnant population is not known.

The aim of the present investigation was to study the incidence, aetiology and prognosis of fetal cerebral ventricular dilatation in a routine ultrasound screening programme.

Methods

From 1 October 1980 to the end of October 1981 the following screening programme was undertaken at the Department of Obstetrics and Gynaecology, University Hospital of Lund: every pregnant woman was offered ultrasound examination twice during her pregnancy. The first examination was made at 17 weeks gestation and the second at 32 weeks. All but five of the 3125 patients participated in the study.

The ultrasound examinations were performed with a (Hitachi EBU 22) ultrasound real-time scanner with a 3.5 MHz transducer. The examinations were made by trained midwives under the supervision of one member of the

research team (C.J.). For later documentation polaroid photographs were taken. The ventricular system was assessed as described by Garrett *et al.* (1975). The width of the lateral ventricle (LVW) was measured at the level of the midbody from the middle of the midline echo to the first strong echo of the lateral wall and the hemisphere width (HW) at the same level from the middle of the midline echo to the first strong echo of the inner table of the skull. Ventricular dilatation was diagnosed by increase of the LVW/HW index according to previously established normal criteria (Denkhaus & Winsberg 1979; Morgan *et al.* 1979; Johnson *et al.* 1980; Fiske *et al.* 1981; Levene 1981).

After birth all positive findings were confirmed by another ultrasound examination. All infants participated in the Child Health Care Clinic programme which includes head circumference measurements and routine developmental screening. Their records were examined again for signs of cranial abnormalities appearing within the first 3 months of age. The registers of the Child Health Care Centres and the Department of Paediatrics have been investigated for any case of congenital hydrocephalus diagnosed in the postnatal period.

Results

In the 3120 pregnant women studied dilatation of the brain ventricles was discovered in five fetuses (0.16%). Table 1 presents a summary of the clinical data.

Patient no. 1

This 29-year-old primigravida had normal findings with ultrasound at 17 weeks gestation.

Table 1. Clinical data at birth, diagnosis, delivery method and follow-up of four liveborn infants and one fetus with dilatation of brain ventricles

Patient no.	Gestational age (weeks)		Method of delivery	Sex	Birth-weight (g)	Head circumference (cm)	Diagnosis	Follow-up
	At diagnosis	At delivery						
1	35	39	Caesarean section	F	3020	37	Lumbosacral myelomeningocele	Lower legs flaccid paresis Bladder paresis Slight psychomotor retardation
2	34	42	Vertex vaginal	M	3820	35	Lumbosacral myelomeningocele	Bladder paresis Normal psychomotor development
3	32	38	Vertex vaginal	M	3000	36	Coxsackie B1-virus infection	No paresis Normal psychomotor development
4	32	37	Caesarean section	M	2390	34.5	Meningo-encephalocele	No paresis Slight psychomotor retardation
5	23	23	Hysterotomy	—	—	—	Thanatophoric dwarfism	—

The second scan at 32 weeks showed the fetal biparietal diameter (BPD) to be smaller than expected. Consequently a further examination

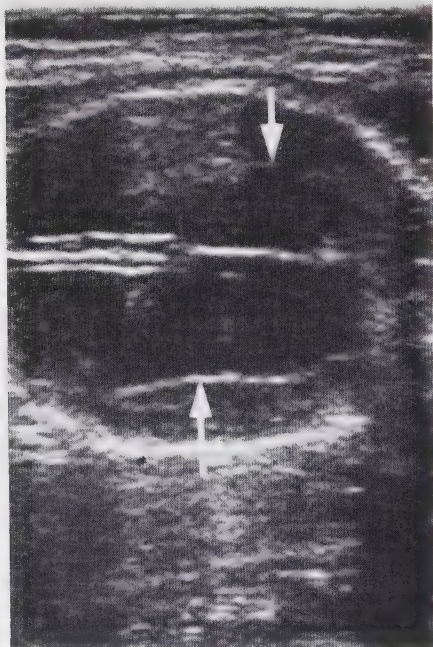


Fig. 1. Myelomeningocele (patient no. 1). Bodies of lateral ventricles (arrows) in transverse section.

was made at 35 weeks and the BPD indicated an accelerated enlargement of the fetal head. A dilatation of the brain ventricular system affecting the lateral ventricles and the third ventricle was found (Fig. 1). A thorough examination of the fetal spine was not conclusive as the fetus was a breech presentation. Further weekly examinations showed that the BPD increased normally but it was smaller than expected for the gestational age. At 39 weeks a female infant was delivered by caesarean section, for maternal reasons. The baby had a lumbosacral myelomeningocele and the large fontanelle was slightly bulging. Hip flexion and adduction were normal but the hips were dislocated, the knee joints were incapable of either extension or flexion and there was right pes equinovarus. On the first day of life the myelomeningocele was closed surgically. The brain ventricular dilatation, followed by B-mode echoencephalography, showed increasing LVW/HW indices and at 17 days of age the baby was given a ventriculoperitoneal shunt. At 1 year of age the child could sit but had flaccid paresis of the lower legs. Her psychomotor development was only slightly retarded.

Patient no. 2

This 31-year-old primigravida had normal findings with ultrasound at 17 weeks gestation.

The scan at 32 weeks showed an oval skull with a BPD that was smaller than expected for the gestational age. Further examination 2 weeks later showed a normal BPD and abdominal diameter, but a moderate symmetrical brain ventricle dilatation was seen; the spine was normal. At 42 weeks a male infant was born after a spontaneous vertex delivery. At birth a very small skin-covered lumbosacral myelomeningocele was found and treated surgically on the first day of life. There was no bone defect and the lower extremities functioned normally but the baby had bladder paresis.

On the third day of life B-mode echoencephalography demonstrated a moderate dilatation of the lateral ventricle with a LVW/HW index of 21/44 on the right side and 25/44 on the left side. There was progressive dilatation during the first week of life. At 12 days of age the boy was given a ventriculoperitoneal shunt and at 8 months he had developed normally but with persistent bladder paresis.

Patient no. 3

This multiparous 26-year-old had a normal ultrasound scan at 17 weeks. A scan at 32 weeks showed a considerable asymmetrical dilatation of the brain ventricles with a LVW/HW index 0.33 on the left side and 0.66 on the right side. Weekly increase in BPD was normal and a spontaneous vertex delivery at 38 weeks resulted in the birth of a male infant without any external malformations. During the first week of life poor sucking necessitated tube feeding and the baby was slightly hypertonic. Supraventricular extrasystoles were seen on the electrocardiogram. Bacterial cultures and serology were normal. In the cerebrospinal fluid there was slight pleocytosis (23 monocyte $\times 10^6/l$). Coxsackie B1 virus was isolated and Coxsackie B virus meningitis and myocarditis was diagnosed.

On the second day of life the LVW/HW index was 20/42 on the right side and 16/42 on the left side; at 6 weeks of age they were 19/42 and 16/42. Cranial computerized tomography at 5 days of age also showed asymmetrical ventricular dilatation of both temporal horns.

At follow-up the head circumference had increased normally but continuously on +2SD in relation to his age. The psychomotor developments was normal at 1 year of age. Congenital hydrocephalus in this baby was probably related to intrauterine Coxsackie B1-virus infection.

Patient no. 4

In this 28-year-old primigravida the ultrasound scan at 17 weeks showed a normal BPD but the abdominal diameter was 5 mm smaller than expected. At 32 weeks the BPD was 93 mm but the abdominal diameter was only 85 mm, in addition there was a marked asymmetrical dilatation of the lateral ventricles. Adjacent to the fetal face a cystic structure with solid areas was observed compatible with the diagnosis of frontal meningoencephalocele (Fig. 2). A male infant was delivered by caesarean section at 37 weeks, the

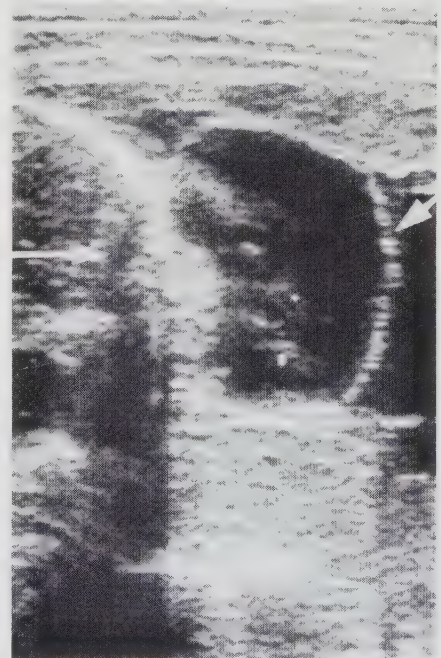


Fig. 2. Meningoencephalocele (patient no. 4). Fetal face in profile. Fetal orbit (arrow).

baby had a 9 $\times$ 6 $\times$ 6 cm cystic translucent swelling protruding from the glabella covering half the nose and both eyes. Cranial X-ray examination revealed a small bone defect in the glabella and B-mode echoencephalography showed a large bilateral dilatation of the brain ventricular system.

On the second day of life the encephalocele was removed. Cranial computerized tomography on the 12th day of life confirmed the findings of the ultrasound examinations.

At 10 weeks, because of the increasing head circumference, the baby was given a ventriculo-

peritoneal shunt. Subsequently, the head circumference grew normally and at 8 months the baby had only a slight psychomotor retardation.

Patient no. 5

In this 35-year-old multiparous patient a diagnostic amniocentesis at 17 weeks revealed a normal chromosomal pattern and α -fetoprotein in the amniotic fluid. At 23 weeks the patient had a second ultrasound examination, because of the abnormally large fundal height. The fetal head had an irregular contour with a large prominence in the neck. There was marked dilatation of the lateral ventricles but the spine was normal (Fig. 3). The fetus was also growth retarded. A legal abortion was performed. The fetus had a grossly deformed head, short extremities and a distended abdomen, thanatophoric dwarfism with clover-leaf malformation of the skull.

Re-examination of files for the Child Health Care Centers and the Department of Paediatrics revealed no further case of congenital hydrocephalus in the infant population up to 3 months of age.

Discussion

In the present study of 3120 pregnancies we found five fetuses with dilatation of the cerebral ventricles: an incidence of 0.16%. This rate is comparable with previous population studies of infantile hydrocephalus which suggest an incidence of 0.14% (Lagergren 1981). But whereas postnatal studies (Lorber 1961; Hill & Volpe 1981) may include both antenatal and postnatal cases of hydrocephalus the present investigation only includes true congenital dilatation of brain ventricles. An additional potential benefit with the present screening system is the possibility of diagnosing major congenital abnormalities early in pregnancy in time to allow a legal termination of the pregnancy.

In our fetuses with myelomeningocele the BPD was smaller than the average for the gestational age. This has been described as a typical finding (Little & Campbell 1980). Furthermore the growth of the BPD was normal indicating a normal pressure hydrocephalus (Hill & Volpe 1981), so that normal labour and delivery at term could have been expected.

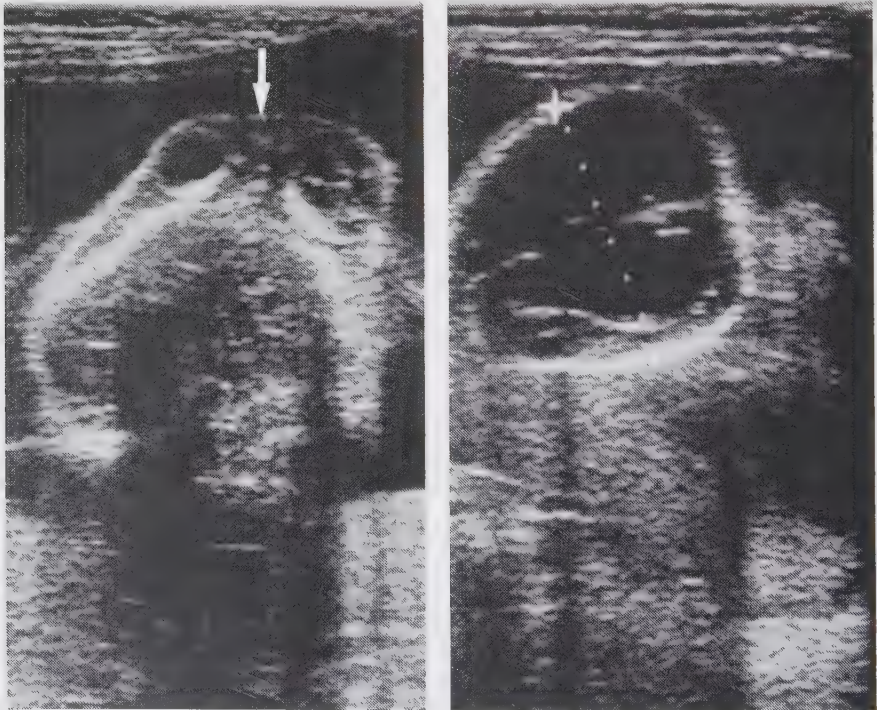


Fig. 3. Thanatophoric dwarfism (patient no. 5). Transverse section of the fetal head. At the level of the brain stem a large prominence in the neck (left). Irregular contour of the skull and a marked dilatation of the lateral ventricles (right).

In patient no. 3 the BPD was almost normal and without accelerated growth and the child was born spontaneously at 38 weeks. This is an additional case report of normal pressure hydrocephalus due to a viral infection and with no myelomeningocele.

Antenatal diagnosis by ultrasound of occipital encephalocele has been reported by others (Miskin *et al.* 1978; Sabbagha *et al.* 1978). Patient no. 4 represents a case of frontal encephalocele. This fetus had a high pressure hydrocephalus as indicated by an accelerated increase of BPD in comparison to abdominal diameter. The size of the meningoencephalocele was considered to be obstructive and consequently caesarean section was performed at 37 weeks.

There is no doubt that all patients with severe intrauterine hydrocephalus should be diagnosed early so that the pregnancy can be terminated (Freeman *et al.* 1977); in the present series this was possible in only one patient. In the latter part of pregnancy the intracranial structures are easier to visualize and there is still the potential benefit of deciding the best time and mode of delivery and arranging rapid follow-up and treatment after birth.

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III

Brain ventricle dilatation in severely ill newborns.

**A clinicopathological ultrasonographic study of
possible causes in the ante-, intra- and neonatal period**

C. Jørgensen, N.W. Svenningsen & I. Ingemarsson



Summary

During a 2 8/12 year period, 27 infants with gestational age from 25 to 42 weeks developing progressive brain ventricle dilatation during the neonatal period of life were studied for possible causes during the antepartum, intrapartum and neonatal period. Most infants were born in difficult deliveries or with perinatal complications. Thus 20 of 27 infants needed respirator therapy. The diagnosis of ventricle dilatation was made by ultrasound examinations in most cases confirmed by computerized tomography (CT) or autopsy. Antenatal ultrasound examination had been made in 25 of 27 cases showing no signs of ventricle dilatation before delivery i.e. in 16 infants one week, in 3 infants six to nine weeks before birth and in 6 infants at 17th gestational week. In 20 infants the ventricle dilatation was possibly posthemorrhagic while in 7 infants no definite sign of intracranial hemorrhage was found. In 14 survivors 4 had severe and 6 moderate cerebral palsy indicating the prognostic severity of early ventricle dilatation after perinatal complications. The single most important perinatal factor correlated with progressive ventricle dilatation and poor outcome was asphyxia. Our observations confirm the usefulness of routine ultrasound examinations of newborn infants after complicated perinatal events.

Introduction

Ultrasound examination of the neonatal skull has become an established supplement to the clinical evaluation of the newborn infant. Detailed information of the intracranial anatomy is obtained (1, 2). Measurement of the width of the lateral ventricle has been used to discover dilatation of the ventricular system, both in the antenatal and the newborn period of life (3, 4, 5, 6, 7).

A major area for application of this technique is early noninvasive diagnosis of intracranial hemorrhage in the newborn.

Various systems for grading of the hemorrhages have been described and attempts been made to correlate the degree of hemorrhage and later psychomotor development (8, 9, 10, 11, 12, 13, 14, 15).

Improved obstetrical and neonatal management has increased the survival rate for newborn infants. Nevertheless intracranial hemorrhage may occur especially among preterm infants and term infants with severe delivery complications. Therefore it is of great importance to evaluate those perinatal conditions which may influence the outcome (16, 17, 18). It is equally important to evaluate the intracranial ultrasound diagnosis in relation to the development of the infant.

In the present study we have electively followed those infants who developed progressive brain ventricular dilatation diagnosed by ultrasound examination during the neonatal period.

We have decided to use the ventricle enlargement as the primary selection criterion and not the degree of hemorrhage, since existing systems of grading the extent of hemorrhages still have limitations in correlations of structure (neuroanatomy by ultrasound) with function (outcome of patient) (19). The ante-, intra- and neonatal periods have been analyzed for possible etiological causes in order to evaluate the clinicopathological - ultrasonographic (ventricle dilatation) correlation with later outcome.

Method and Material

Ultrasound examination of the skull has been used at the Neonatal Intensive Care Unit (NICU) when gross abnormal clinical findings occurred (i.e. signs of central nervous system disorders such as bulging fontanel, abnormal head circumference, hypo- or hypertonus and seizures) and only in a few cases as a routine procedure. From November 1980 through June 1983 dilatation of the lateral ventricles was diagnosed in 27 infants.

A Hitachi EBU 22 linear array real-time ultrasound scanner with a 3.5 MHz transducer was used. Later even a sector scanner Diasonic DS 20 with a 5.0 or 6.0 MHz transducer was available.

Measurements of the lateral ventricles and calculation of ventricular/brain index in relation to degree of maturity at birth and postnatal age were performed as described by Levene 1981 (22). Infants with ventricular/hemisphere-index of mean + 2 SD for the gestational age were included. In the present study values for the index varied from 0,38 to 0,81.

Other diagnoses as intracranial hemorrhage or signs of cerebral atrophy with a wide interhemisphere fissure were also registered. In this material no attempt was made to classify the degree of hemorrhage. Postmortem examination was performed in all children who died except one. Cranial computerized tomography and/or cerebrospinal fluid (CSF) cytology or total protein examinations were used as adjuvant diagnostic tools in 20 of the 27 infants. The occurrence of sidero- and erythrophages in the CSF cytology examination was used as one indicator of intracranial bleeding (23).

In this study evaluation of neurodevelopment was made at 1 1/2 - 3 years of age according to Milani-Comparetti developmental score (24). Light CP injury (cerebral palsy syndrome) include children with only slight motor retardation (i.e. 2 months delay of development taking into consideration conceptional age at birth), moderate CP hemi- or diplegia in combination with moderate motor retardation (i.e. 3 months delay of development) and severe CP tetraplegia, microcephaly and severe psychomotor retardation.

Statistical evaluation with χ^2 -test was applied when comparing the survivors with non-surviving infants.

Results

In 27 infants (11 girls and 16 boys) dilatation of the lateral ventricles developed during their hospitalization in the NICU. Of these 15 were inborns at the Department of Obstetrics in Lund and 12 were outborns from other delivery departments in the regional area of Lund. Gestational age at birth varied from 25 to 42 weeks with a mean of 32 6/7 weeks of gestation. Nearly all, i.e. 25 of 27 infants, had been examined by antenatal ultrasound with no sign of ventricular dilatation. In 16 this examination was made less than 1 week before the delivery, in 3 cases at week 32, i.e. 6 to 9 weeks before delivery, and in 6 only an early examination in gestational week 17 was made, i.e. 10 to 23 weeks before delivery. The remaining 2 children were both examined on the 4th day of life with a normal result concerning the ventricular width. Table I. shows gestational age and weight at delivery for the 27 infants.

Table I. Gestational age and weight at delivery.

gestational age		25-30 weeks	31-35 weeks	> 35 weeks

sex	female	3	3	5
	male	7	5	4
birthweight		< 1000g	1000-1500g	1500-2500g > 2500g

		10	3	9 5

In the 27 infants 125 ultrasound scannings were performed (mean 4,6/patient). Three infants were examined only once, but the ultrasound diagnosis was verified (2 by postmortem examination and 1 by computer tomography = CT). Another two infants had only 2 examinations, but even in these CT-examination confirmed the diagnosis. In 16 cases besides ultrasound also CT examinations were made after varying intervals from 1 to 144 days. However, in all 16 cases

ventricular dilatation was confirmed by the CT examination.

Discrepancy between ultrasound and autopsy was found in one case: ultrasound showed atrophy on day 34, confirmed at autopsy on day 43, but in addition subdural hemorrhages were found, which had not been diagnosed at ultrasound examinations.

Twenty of the infants had their first examination before or on the 10th day of life, six after 15 to 28 days and one after 81 days of life. This baby born in gestational week 30 had a quite uncomplicated development until 2 1/2 months of age. At that time weight gain stopped and the growth of head circumference decreased. Ultrasound examination showed cerebral atrophy, as did autopsy made when the baby died at 140 days of age.

In 7 of the 27 infants only ventricular dilatation were demonstrated, while 4 also had a wide interhemisphere fissure indicating cerebral atrophy. In 3 of these 4 this was also found at the postmortem examination. The width of the lateral ventricles in these 4 cases of atrophy was within +2 SD, whereas the other 23 infants had lateral ventricles above +2 SD. The ventricle/hemisphere index being above +2 SD in these 4 infants was due to a relative decreased hemisphere width.

Post hemorrhagic ventricular dilatation.

The findings at ultrasound examinations indicated that in 20 cases the ventricular dilatation was possibly related to intracranial (peri/intraventricular) hemorrhages. Furthermore 15 of these 20 infants were also examined with CT-scans, lumbar puncture with CSF cytology examination and/or postmortem examinations. In all 15 these investigations confirmed the findings at the ultrasound examination.

In 5 of the 20 cases the ultrasound diagnosis of intracranial hemorrhage could not be verified, probably owing to a time delay

between ultrasound and the other examinations. However brain ventricle dilatation was confirmed in all.

Rate of mortality and handicap.

The first year mortality rate was high since 13 infants died between 12 and 330 days of age with a mean of 103 days. In the 14 survivors 3 had light, 6 moderate and 4 severe cerebral palsy syndrome (CP). Ventriculo-peritoneal shunt operation was made in 6 infants during the NICU hospitalization. One developed sign of increasing intracranial pressure not until 7 months of age and was then shuntoperated. There was no correlation between outcome and gestational age or birth weight in this selected material (Table II).

One child had a normal neurodevelopment in spite of pronounced ventricular dilatation with an index of 0.70 at one month of age. This child had a peri/intraventricular hemorrhage but it was only demonstrated by ultrasound scanning (Fig 1). At one year of age the index had decreased to 0.42.

Outcome in relation to pregnancy complications.

As shown in Table II the rate and degree of severity of the handicap in infants with brain ventricle dilatation was almost equal in the different gestational age and birth weight groups. In this selected material of infants with brain ventricle dilatation most pregnancies were complicated, i.e. in 17 of 27 abruptio placentae, preeclampsia and intrauterine growth retardation being the most frequent diagnoses. In infants born in the 25th to 30th weeks of gestation all deaths were related to prematurity in combination with other pregnancy or delivery complications. In contrast prematurity alone and without such major complications was present in 2 infants with only moderate CP motorhandicap and in 1 with normal development.

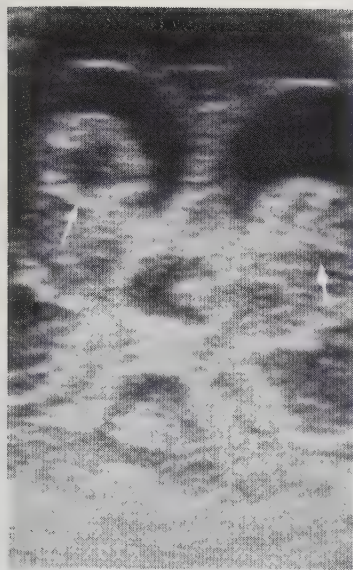


Fig. 1. Shows a coronal scanning through the fontanel (linear 3.5 MHz transducer) with bilateral peri/intraventricular hemorrhages (arrows) and ventricular dilatation in the infant with normal neurodevelopment.

Table II. Outcome correlated to gestational age and birth weight, in infants with brain ventricle dilatation.

	25-30 weeks	31-35 weeks	> 35 weeks	Total
Death	5	4	4	13
Severe CP	1	1	2	4
Moderate CP	2	2	2	6
Light CP	1	1	1	3
Healthy	1	0	0	1
	10	8	9	27

	< 1000g	1000-1500g	1501-2000g	2001-2500g	> 2500g
Death	5	1	2	3	2
Severe CP	2	0	0	1	1
Moderate CP	2	1	0	2	1
Light CP	0	1	1	0	1
Healthy	1	0	0	0	0
	10	3	3	6	5

Even in the other gestational age groups the pregnancy complications were more frequently found among the dead or severely handicapped infants (7/11). In the remaining 6 cases with light to moderate CP, 3 had a normal pregnancy.

Outcome and delivery.

The method of delivery was Cesarian Section (CS) in 21 and only 6 infants were born vaginally, i.e. one in group 25-30 weeks and 31-35 weeks respectively, and four in the group >35 weeks. Indications for CS were in all cases pregnancy or delivery complications as mentioned above.

Table III shows the Apgar-score at 1 and 5 minutes. Fourteen (52%) had a 1 minute Apgar below 4 and 12 (44%) a 5 minutes Apgar below 7. Of those 13 who died 10 (77%) had a 1 minute Apgar below 4 and 9 (69%) a 5 minutes Apgar below 7. Furthermore 10/14 (71%) infants with 1 minute Apgar below 4 and 9/12 (75%) with 5 minute Apgar below 7 did not survive.

Ante- and intrapartum CTG (cardiotocography) was registered in 21 cases. In 6 no CTG was done because of an unexpected complication followed by emergency CS. The CTG was considered as either + or - ominous. The definition of ominous being: Fetal heart rate patterns demonstrated silent pattern and/or late decelerations or complicated variable decelerations or pronounced combined decelerations (25,26). Thirteen CTG registrations showed + ominous and the remaining 8 - ominous. Seven of the 13 with signs of intrauterine asphyxia died compared with 4 with normal CTG, or nearly the same percentage.

Outcome correlated to seizures and pathological EEG.

Seizures occurred more often among infants above 30 weeks of gestation (13/17 or 76%) than in those below 31 weeks (4/10 or 40%). In

the group of 17 infants with seizures, 10 had 1 minute Apgar below 4 and 9 a 5 minute Apgar below 7. In the group of 13 deaths 10 occurred among infants with pathological EEG and seizures.

Table III. Outcome correlated to Apgar-score at 1 and 5 minutes.

	25-30 weeks	31-35 weeks	> 35 weeks	Total no
< 4 at 1 min.	5	3	6	14
< 7 at 5 min.	4	2	6	12

	< 4 at 1 min.	< 7 at 5 min.	Total no
Death	10	9	13
Severe CP	1	1	4
Moderate CP	2	1	6
Light CP	0	0	3
Healthy	1	1	1
	14	12	27

Repeated EEG registrations were made in 16 of the 17 infants with seizures. The electrical activity remained unchanged pathological in 13, showed a change from severe to light pathological in 2 and to normal in 1. Only 5 of 27 had a normal initial EEG. At the follow-up one of these is healthy, 3 have moderate to severe CP and 1 died from pulmonary complications.

Outcome and pulmonary complications.

Twenty of the 27 infants needed respirator treatment for more than 1 week. All 13 who died were in this group. In the group 25-30 weeks all had pulmonary complications such as hyaline membrane disease, bronchopulmonary dysplasia or pneumothorax. Even in the

group 31-35 weeks 7 of 8 had pulmonary problems. In the group > 35 weeks 2 infants had transient tachypnoea but the others had no problems.

In this material 6 of 7 infants with pneumothorax also got an intracranial hemorrhage.

A comparison of perinatal data between the 14 survivors and the 13 who died is shown in Table IV. The infants who died had more pregnancy complications, asphyxia and low Apgar score at delivery ($p < 0,05$). They more often developed seizures and pathological EEG and all needed prolonged respirator treatment ($p < 0,01$).

Table IV. Clinical data on survivors and non-survivors with cerebral ventricular dilatation.

	Death	Survivors	χ^2 -test
Number of children	13	14	P-value
Gestational age	32,6 w	33 w	ns
Mean birth weight	1742g	1833g	ns
Pregnancy complications*	10	7	$\chi^2=2,10$ ns
Ante/intrapartum asphyxia	7	6	$\chi^2=0,33$ ns
Apgar < 4 at 1 min.	10	4	$\chi^2=6,31$ $p < 0,05$
Apgar < 7 at 5 min.	9	3	$\chi^2=6,24$ $p < 0,05$
Seizures	10	7	$\chi^2=2,10$ ns
Pathological EEG	10	7	$\chi^2=2,10$ ns
Pulmonary complications	10	8	$\chi^2=1,19$ ns
Respirator > 1 week	13	7	$\chi^2=8,78$ $p < 0,01$
Peri/intraventricular hemorrhage	10	10	ns

* i.e. abruptio placentae, preeclampsia, intrauterine growth retardation etc.

Discussion

The aim of the present investigation was not to get incidence, but to analyse the pregnancy, delivery and postnatal period for possible causes of intracranial injuries and any consequent deleterious effect from progressive ventricle dilatation.

The most common reason for brain ventricle enlargement during the neonatal period is posthemorrhagic (10, 20, 21, 22, 27). In this study at least 20 children had a possible intracranial hemorrhage as the background for dilatation of the ventricles. Since all ultrasound examinations were made only after abnormal clinical findings had developed, dating of the hemorrhage and the ventricular dilatation was not possible.

Furthermore the present study includes a selected group of severely ill newborns, which may be the reason for the nearly equal size of the gestational age groups. It does not reflect the real distribution of infants with intracranial hemorrhage but without ventricular enlargement.

Our results demonstrate as others (14, 28) a good concordance between ultrasound findings, CT-scans and postmortem examinations in spite of rather long time interval in some cases.

Allan et al (10) and Hill and Volpe (20) have reported a frequent development of ventricle dilatation in infants with pronounced hemorrhage. They propose that the poor outcome of these infants is related to the size of the intraparenchymal hemorrhage and that the ventricle dilatation is merely a symptom. They also found that all infants with severe and large intraventricular hemorrhages later developed ventricular dilatation. This is one possible cause in several infants even in our study considering the very poor outcome in most of our 27 infants.

Still it is important to realize that the pathophysiological effects from enlarged ventricles may be quite different depending on the degree of elevation of intraventricular pressure. Ventriculomegaly ex vacuo may on the other hand imply loss of cerebral tissue and consequently more severe longterm disturbances (19).

In 7 infants or 26% the ventricular dilatation was progressive in the present study. Six of these infants got a ventriculoperitoneal shunt during the first months of life. One developed signs of hydrocephalus late and was shuntoperated at an age of 7 months. Four of the shunt operated infants have survived.

Asphyxia has been considered an important etiological factor for formation of peri/intraventricular hemorrhage (8, 15, 18, 21). It has been suggested that impairment of the cerebrovascular autoregulation may imply vasoconstriction and periventricular leukomalacia or vasodilatation with overperfusion in the subependymal tissue in both instances with a result of subsequent hemorrhage. Some authors have reported a significant correlation between low Apgar score and peri/intraventricular hemorrhage (2,5), while others have not (11). Our results demonstrate a significant correlation between low Apgar score and outcome (see Table III and IV).

Complications during pregnancy and delivery were frequently leading to ante- or intrapartum asphyxia most clearly seen in the age group 25-30 weeks. As soon as the extreme prematurity is combined with asphyxia the outcome is very poor.

During the neonatal period asphyxial episodes with cardiovascular collapse may contribute to development of hemorrhage. Thus we found that 6/7 of infants with pneumothorax developed IVH as also described by others (30).

Seizures were most frequently registered among infants above 30 weeks of gestation. It is well known that the convulsive pattern of premature and fullterm newborn infants differs from that in older

individuals (29). In one study (31) we have found that in infants without clinical seizures, changes in the electrical activity characteristic for seizures (silent seizures) may be found by continuous surveillance with cerebral function monitor or longterm EEG registration. Seizures - overt or silent - may be an additional etiological factor for later neurological handicaps in infants with brain ventricle dilatation in early life (32).

The possibility of antenatal cerebral damage in some infants in the present study cannot be excluded, especially in infants with intrauterine growth retardation (33). Still, in 16 of 27 infants the antenatal ultrasound examinations made within the last week before delivery did not show enlargement of the ventricles.

In conclusion our study demonstrates a very poor outcome for infants with progressive ventricle dilatation after a complicated perinatal period. Ante- and neonatal asphyxia seems to be the single most important factor for development of perinatal intracranial damages. Prevention of asphyxia, i.e. early intervention in deliveries, especially in preterm infants, with signs of asphyxia in CTG and alert neonatal care adjusted to avoid episodes of post-natal asphyxia, may improve the overall outcome for the infant.

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IV

The advantage of antenatal diagnosis of intestinal and urinary tract malformations

C. M. Kullendorff, L. T. Larsson & C. Jörgensen

The advantage of antenatal diagnosis of intestinal and urinary tract malformations

C. M. KULLENDORFF, L. T. LARSSON & C. JÖRGENSEN *Departments of Pediatric Surgery and Obstetrics and Gynaecology, University Hospital, S-221 85 Lund, Sweden*

Summary. In a 24-month prospective screening programme 6020 pregnant women were examined with diagnostic ultrasound at 17 and 32 weeks gestation. In a total of 23 (0.38%) abnormalities, four cases of urinary tract and two of intestinal tract abnormalities were discovered. The antenatal diagnosis influenced the management of these disorders both before and after birth.

Congenital anomalies that affect the newborn are often surgically correctable (Hobbins *et al.* 1979; Canty *et al.* 1982), and reduction in morbidity and mortality may be achieved by earlier, more precise diagnoses and prompt correction. The antenatal recognition of surgical disorders by ultrasound examination has brought changes in the perinatal and postnatal management and improved the prognosis for the infant (Touloukian & Hobbins 1980; Harrison *et al.* 1981).

The present study was designed to assess the frequency of antenatal diagnosis of intestinal or urinary tract malformations and the influence on the management.

Patients and methods

During the 24-month period October 1980—October 1982 all pregnant women in the district of Lund (6020) were routinely examined by ultrasound in the 17th and 32nd week of gestation.

The ultrasound examinations were performed by trained midwives under the supervision of specially trained doctors, using an ultrasound realtime scanner (Hitachi EBU 22) with a 3.5 MHz transducer. For documentation polaroid photographs were taken and video-recordings were made. All anomalies were noted and further investigated.

Results

In the 6020 pregnant women examined a total of 23 (0.38%) fetal anomalies were diagnosed. Nine

pregnancies with major fetal abnormalities were found at week 17 and they were legally terminated.

Six fetuses had abnormalities of the central nervous system (Jørgensen *et al.* 1983), one had Potter's syndrome and one hydrothorax and hypoplastic lungs. The clinical course of the other six fetuses is the subject of this paper: four had anomalies of the urinary tract and two had intestinal tract anomalies.

Patient no. 1

This 30-year-old woman was in her first pregnancy. An ultrasound scan at week 17 revealed twins and was repeated every 3 weeks to monitor growth. At week 32 both twins demonstrated a cystic structure in the region of the right kidney. Further examinations confirmed these findings indicating dilatation of the right renal pelvis, most marked in twin I. Rupture of the membranes occurred at 32 weeks but it was possible to stop the labour with terbutalin until 34 weeks. Caesarean section was performed because of prematurity and breech presentation in twin I, who weighed 2400 g. Both babies were male and appeared to be monozygotic. Post-partum ultrasound revealed that dilatation of the renal pelvis in twin I was slightly reduced but still considerable. In twin II the smaller dilatation remained unchanged. Twin I had an intravenous pyelogram (IVP) at 3 weeks of age. There was no

excretion from the right kidney. A retrograde pyelogram showed a hydronephrosis with an obstruction at the uretero-pelvic junction, and at 5 weeks the infant had an Anderson-Hynes dismembered pyeloplasty and nephrostomy. At operation preserved renal parenchyma was noticed. The baby was discharged from hospital 6 days after the operation and the nephrostomy was closed 5 weeks later. Twin II also had a right sided hydronephrosis but IVP showed good excretion from the right kidney. The hydronephrosis in this baby was also caused by an obstruction in the uretero-pelvic junction but operation was not indicated. This healthy child is being followed up with repeated IVP.

Patient no. 2

This 26-year-old woman was in her second pregnancy. An ultrasound scan at week 17 was normal, but at week 32 a slight polyhydramnios was noticed. Further examination revealed a cystic structure in connection with the left kidney suggesting dilatation of the left renal pelvis. A male infant weighing 3960 g was delivered by caesarean section at 43 weeks because of prolonged labour. Postpartum ultrasound confirmed the prenatal finding, and investigations including IVP, urethrocystography and cystoscopy showed a distal ureteric stenosis. At the age of 6 months an ureteroneocystostomy by the technique of Politano-Leadbetter was performed. Follow-up radiological examination showed reduction of dilatation of the urinary tract in an otherwise healthy boy.

Patient no. 3

This 24-year-old primigravida had a normal ultrasound at 17 weeks gestation. At 32 weeks a large cystic structure was seen in the region of the right kidney of the fetus, with another smaller cystic structure situated medially. The left kidney was normal. The outcome of a spontaneous vertex delivery at 39 weeks was a male infant weighing 3950 g with no external malformations. Within 12 h radiological investigations were performed. An IVP showed no excretion from the right kidney. Investigations of the gastrointestinal tract aroused suspicion of an atresia in the sigmoid colon, and after preoperative rehydration laparotomy was performed at less than 24 h of age. At operation two colonic

atresias were found distally in the sigmoid colon. The affected parts were resected and one end to end anastomosis was performed without colostomy. Palpation during operation revealed a right polycystic kidney. The child was discharged from hospital in good condition 18 days after surgery and follow-up showed a normal colonic anastomosis. The polycystic kidney has shown no function and nephrectomy is planned.

Patient no. 4

This 30-year-old multigravida had a normal ultrasound scan at 17 weeks. Rupture of the membranes occurred at 22 weeks but with only terbutalin treatment the pregnancy continued until 33 weeks. She was regularly followed with ultrasound. At 29 weeks the fetus showed a dilatation of the stomach and the proximal part of duodenum, indicating atresia of the duodenum. Caesarean section was performed in the 33rd week of pregnancy because of prematurity in combination with breech presentation. A boy weighing 1800 g was delivered. Radiological examination confirmed the diagnosis of duodenal atresia, but because of an infection with *Escherichia coli* the baby was not referred to the pediatric surgical unit until the second day of life. The baby was discharged from hospital 5 weeks after operation weighing 2510 g, and follow-up studies showed a well functioning anastomosis and a normally developing infant.

Patient no. 5

This 20 year-old woman in her second pregnancy had a normal ultrasound scan at 17 weeks. She was referred for further examination at 31 weeks because of suspected intrauterine growth retardation. Another examination at 33 weeks showed the fetus to be smaller than expected but with normal growth. In addition it was found that the fetus had a defect in the anterior abdominal wall with the entire bowel floating free in the amniotic fluid. These findings indicated gastroschisis and contact with a pediatric surgeon was established. At 38 weeks the membranes ruptured and an emergency caesarean section resulted in the birth of a female baby weighing about 2300 g who showed great vitality. Meanwhile the neighbouring operating theatre was completely prepared for immediate operation of the baby. Within 5 min the operation started and 20 min after the birth

primary skin closure of the gastroschisis was performed. Fascial closure was not possible. Mechanical ventilation was maintained for the first 15 h postoperatively because of the highly increased intra-abdominal pressure. The clinical course was then uneventful. The baby was discharged from the hospital earlier than usual and has remained well.

Patient no. 6

This 29-year-old primigravida had a normal ultrasound scan at 17 weeks. At 32 weeks a marked distention of the fetal bladder was observed; the kidneys and renal pelvis were normal but the ureters were slightly dilated. The fetus was a female. The quantity of amniotic fluid was normal. The distention of the bladder did not increase but was estimated at 200 ml, and at week 33 a percutaneous bladder puncture was performed with aspiration of about 100 ml urine. After 16 days the bladder had refilled to the pre-operative size indicating a normal renal function. It was planned to introduce a percutaneous bladder-amniotic fluid shunt but unexpected intra-uterine fetal death occurred at 36 weeks. Aseptic autopsy showed no sign of intrauterine infection and no pathological conditions apart from a dilated urinary bladder. The cause of death could not be explained.

Discussion

Ultrasound examination in pregnancy is a safe and valuable method of fetal surveillance; long-term studies have shown that it has no deleterious effects on the growing infant (Scheidt *et al.* 1978). Many fetal anomalies may be diagnosed (Garrett *et al.* 1975; Kurjak *et al.* 1980), and advanced knowledge of them may influence both obstetrical and surgical management (Björnsthål *et al.* 1983).

The present study is particularly valuable in giving accurate incidence rates for fetal abnormalities of the intestinal and urinary tract. Other published series have been from referral centres, but we excluded all referral cases.

The urinary tract disorders are often not obvious at birth and they may cause relatively few clinical symptoms but they may be dangerous in leading to a stealthy destruction of the kidneys.

In the study one baby with hydronephrosis was operated on within 5 weeks of delivery (Patient no. 1). The ultrasound could not precisely demonstrate the obstruction, but the single cystic

dilatation in the kidney area was highly suggestive of obstruction at the uretero-pelvic junction. After delivery IVP confirmed the obstruction and the antenatal diagnosis was decisive for the early operation. The obstruction was thereby removed and further destruction of the renal parenchyma halted. The antenatal diagnosis was valuable in this patient and also in patient no. 2 with hydronephrosis and hydroureters due to distal ureteric stenosis in a symptom-free baby.

In an infant with polycystic kidney the surgical treatment is elective. In patient no. 3 the affected kidney was palpable at birth. During the immediate operation due to atresia of the colon the right kidney was palpated and found to be enlarged and cystic. The atresia was corrected, but the kidney anomaly was left for further function investigations. The treatment of the polycystic kidney is contentious; resection is most often advocated and can be performed electively.

An intestinal anomaly was revealed in patient no. 4 with a suspicion of a duodenal atresia. The operation was performed 2 days after delivery with a duodeno-jejunostomy. Duodenal atresia often does not become evident clinically before 24–48 h when the baby then starts vomiting after feeding. With prolonged vomiting, the hydration and electrolyte imbalance can cause preoperative problems and further delay. Duodenal atresia is not a life-threatening anomaly in the first hours of life, but advance knowledge can, as in our patient enable proper planning of early correction.

In patient no. 5 with gastroschisis, the neonatal and surgical services were consulted well in advance of estimated delivery. Although the onset of labour was acute, careful preparations could be made for appropriate infant care after delivery. Most of these infants have been born vaginally at term and the malformation discovered at birth. When delivery does not occur in a hospital with a paediatric surgical department, transport of the infant for prompt repair is necessary. The issue of vaginal delivery or elective caesarean section is still debatable. In larger omphaloceles and gastroschisis there is a risk of trauma due to difficult prolonged labour. Section is in our opinion still advantageous, but the increased risk to both mother and child must be assessed in proportion to reduction of neonatal mortality. In this patient the management was regarded optimal. The mother was transported to the surgical department where the section was performed and the child was immediately operated in the adjacent theatre.

Fetal surgery with repair or palliation of some defects may be considered in the future. It is rapidly improving but only a few cases have been attempted in the human fetus. In three areas fetal intervention may be beneficial. One condition is distal urinary tract obstruction resulting in dilatation of the upper urinary tracts and deterioration of the kidneys *in utero*. The female fetus in patient no. 6 with only a dilated urinary bladder was assigned to this group. A splint for permanent intrauterine bladder drainage into the amniotic fluid has recently been described (Berkowitz *et al.* 1982). The other malformations are diaphragmatic hernia and hydrocephalus but they are not discussed in this context.

Maternal ultrasound scanning will probably be regarded as an important factor for the improvement of infant survival. If an anomaly of the intestinal or urinary tract is diagnosed *in utero* we recommend referral of the mother, if necessary so that delivery can take place in a hospital with both neonatal and pediatric surgical facilities.

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Ultrasound diagnosis of fetal malformation in the second trimester. The psychological reactions of the women

C. Jörgensen, N. Uddenberg & I. Ursing

Abstract

During a 3-year period 12 fetal malformations were diagnosed by routine ultrasound scanning in the 17th week of gestation. Semi-structural interviews were arranged in order to improve understanding the women's mental reactions, their decision process, their experience of the abortion and the value of the given information. In 1984, 10 women were interviewed during a 2-month period. The time interval between abortion and interview varied between 6 and 34 months. It was found that all the women who had had an abortion because of fetal malformation experienced a severe psychological trauma. In some cases, the crisis lasted for an extended period of time. In this study, half of the patients had been unable to cope with their personal crisis. Hence, it is of utmost importance that the obstetrician is ready to offer a more active psychological support to these women.

Introduction

Ultrasound scanning has generally become accepted as a valuable contribution to the clinical examination of pregnant women. It has been used routinely at the University Hospital of Lund since 1st October 1980. Every pregnant woman planning to be delivered at the Department of Obstetrics in Lund is offered 2 ultrasound examinations, in the 17th and the 32nd week of gestation, respectively. The primary intention of the 17th week examination is to date the pregnancy, according to measurement of biparietal diameter, and also to detect multiple pregnancies. The 32nd week examination is carried out in order to evaluate fetal growth. In addition, information about fetal malformation is obtained. This study presents the results of interviews with 10 women in whom fetal malformation was discovered in the 17th week of gestation.

The diagnosis of and given information about a malformed fetus will give rise to a crisis reaction. Bowlby (1), Parkers (2) and Cullberg (3) have described 4 stages during a psychological crisis.

1: The shock phase, characterized by denial and repression of the trauma. 2: The phase of yearning and searching, when the feeling of guilt may be the essential problem. 3: The disorganization period, when grieving may give way for a feeling of depression, self-devaluation and even apathy (2). At this stage the woman starts coping with the trauma and may again begin to believe in the future (3). 4: Reorientation is complete when the mother decides either to start a new pregnancy or to be content with the situation.

When fetal malformation has been diagnosed, most patients elect to have the pregnancy terminated. The loss of a desired infant in the second trimester might be compared to perinatal death (4, 5). Kirkely-Best & Kellner (6), studying fetal death in late pregnancy, found the grief reactions of these parents to be similar to those with perinatal loss. It has also been found that suppression of feelings in connection with stillbirth may, in some cases, result in a prolonged psychological symptomatology (3). The process of mourning after an abortion of a malformed fetus in the 17th week of gestation may be complicated since the parents may have only vague perceptions of what they are grieving for (7).

A decisive difference between stillbirth and abortion of a malformed fetus is that in the case of abortion the parents have actively decided to terminate the pregnancy, thereby causing the death of a living fetus. The fact that the fetus was malformed might result in even stronger self-accusations and feelings of guilt (8, 9). It is well known that a legal abortion has profound consequences for the woman's reproductive life (10). The abortion of a malformed fetus might influence this on an even deeper level, and the feeling of failure may increase the woman's wish to prove her reproductive capacity through a new pregnancy (3).

The present study was undertaken to explore the experiences of those parents who, after an ultrasound examination in the second trimester, were informed about a fetal malformation, in order to enable the obstetrician to provide better emotional support.

Patient sample

During a 3-year period, 8422 women underwent a routine ultrasound scan on two occasions during their pregnancy. In the 17th week of gestation, major fetal malformations were found in 12 pregnancies (Table I). All these women chose to terminate the pregnancy. One woman was pregnant twice and had a malformed fetus both times. Another woman had left Sweden for Zimbabwe and thus could not be contacted. Hence, 10 women could take part in the study. The time lapse from abortion to interview varied between 6 and 34 months with a mean of 1 year and 5 months. All women were living with the same partner both at the time of diagnosis and at the time of interview. Their age at the time of the interview varied between 25 and 39 years with a mean of 34 years and 9 months. Four women had a university degree, 3 college education and 3 basic schooling.

Table I. Twelve fetal malformations diagnosed by ultrasound in the 17th week of gestation. (X0 and XX) refers to sex chromosomes

Anencephaly	3
Thanatophoric dwarfism	2
Cystic hygroma of the neck (X0 and XX)	2
Myelomeningocele	1
Meningoencephalocele	1
Conjoined twins	1
Hydrops fetalis (X0)	1
Congenital Finnish type nephrosis	1

	12

One woman was primipara, 4 had one child and 5 more than one. One woman had lost a 3.5-year-old child 6 months prior to the abortion. Post-mortem examinations revealed that this child and the aborted fetus both suffered from renal malformation, i.e. congenital Finnish type nephrosis. Two women had received treatment for fertility problems for a short time, but not immediately prior to this pregnancy.

Method

The routine ultrasound scans are made by specially trained midwives. The midwife usually makes the preliminary diagnosis of fetal malformation and refers the patient to a specially trained obstetrician for confirmation. He then informs the woman. This normally takes place on the same day, but sometimes 1 or 2 days may pass before the patient is examined by the doctor. The midwife will only tell the patient that she has made an observation which requires reexamination by a doctor. The screen of the ultrasound apparatus is always turned towards the patient allowing her to look at the picture. Another control examination is usually carried out a few days later in order to reconfirm the diagnosis. This allows the parents to think over and discuss the situation.

In Sweden, abortion is legally allowed until the end of the 18th week of gestation. To abort at a later stage of pregnancy, permission must be obtained from The National Board of Health, and this may delay the termination by a few days. Termination is carried out by hysterotomy or extra-amniotic Rivanol instillation. The patient is hospitalized 2-6 days, depending on the method used and is asked to return to the department approximately 1 month later for a routine physical examination as well as information about the result of the autopsy. Sick leave is generally 1 month. Photographs of the fetus are always taken for documentation of the diagnosis, and sometimes to show to those parents who did not see the fetus but later feel a need to do so.

In January and February 1984, the 10 women were informed by letter about the present investigation. They were then contacted by telephone to arrange a date for interview. All 10 patients agreed to participate.

The interview was semistructural, i.e. 1) the questions, designed in advance, were asked in a certain order, 2) the woman was allowed to elaborate her answers freely, 3) the interviewer wrote

down the answers during the interview and categorized them by number, according to a previously defined alternative answer-schedule (11, 12). Five interviews took place at the Department of Obstetrics and Gynecology, 5 in the women's homes. A 45- minutes interview was planned but the actual time spent varied between 1-2 hours with a mean of 1.5 hour. The atmosphere was friendly and informal. A total of 21 items was discussed, the following subjects being dealt with: information about the fetal malformation, the woman's decision to elect abortion, her reaction to termination of the pregnancy and the influence on her future reproductive life.

Results

The information as experienced by the women.

Eight (8/10) women considered that they had received sufficient factual information. Two considered the information to be sufficient, but nevertheless felt a need for further discussion. One woman said: "I wanted to put questions, which I knew could never be answered". Three women (3/10) were informed gradually while 7 were informed directly at the ultrasound scanning. Irrespective of the procedure the women were content. Four were accompanied by their husbands when the diagnosis was made and considered this to be very positive and important. Six were alone with the obstetrician when given the information. At the subsequent interview, these women still felt they would not have wished to delay the information until the husband could have been present.

It is well known that during the first stage of a crisis reaction the individual may not be able to understand detailed information (3, 5, 7). This was confirmed by the women in this study: "I was desperate, wanted to disappear, to get out as soon as possible". "I was totally cut off from any more information".

Decision process and experience of abortion.

Only 2 women (2/10), considered the possibility of continuing the pregnancy and all but 1 (9/10) found the decision to have a legal abortion easy. Being told by the doctor that the child would never survive apparently played a very important part in their decision-making. One woman, however, hoping that the child might survive, found it very hard to decide about the abortion.

Seven women said that as a matter of principle they were against abortion, but found it acceptable when the fetus was malformed. When questioned about the possibility of another subsequent termination, 2 answered that they would never go through with it. Seven said that they would do it only if the fetus was malformed, and only 1 woman would consider it even for other reasons. The fact that the women terminated their pregnancies in spite of their attitudes based on principle, indicates that they experienced ethical conflicts. Three women (3/10) described their abortion as a terrible experience, which may suggest psychological conflicts: "I felt awful on the morning before the operation. I felt it was so wrong because I was just as pregnant as before".

The time between diagnosis and operation varied between 1 day and 3 weeks. One woman decided to wait 3 weeks because of a planned holiday: "I would not disappoint my family, nothing could be changed in any case". In 2 cases there was an involuntary delay of 2 and 2.5 weeks respectively, which the women experienced as unacceptable. One of these women described the waiting as one long, terrible nightmare. Several women mentioned that it was intolerable to feel the fetus moving.

It may be advisable, however, to have a delay of a few days between diagnosis and abortion. Four women (4/10) said that it was important that the gap was not too short: "It was necessary to have time to get used to the thought that you are going to lose something".

All patients had an appointment at the Department of Gynecology about 1 month after the abortion for a physical examination and for information about the results of the autopsy of the fetus. Nearly all women felt it was very important to see the same obstetrician. In the present study 4 (4/10) patients were satisfied with the visit, whereas 6 reported that it had only been a physical check-up without emotional support. Four women (4/10) mentioned spontaneously that the time of expected child-birth was emotionally difficult, and that they had felt the need for medical appointment at that time: "I had started to feel better, but at the time when the child was due to have been born, it was all back again".

Psychological reactions.

Most patients (7/10) reported very strong reactions when told the diagnosis, whereas 3 had a more moderate reaction. Half of the women (5/10) had been given some kind of warning before the final diagnosis (increased alfa-fetoprotein in 3 cases, polyhydramnios in 1 and 1 patient had had a previous ultrasound examination showing a fetus too small for gestational age). The reactions of these 5 patients were just as strong as those of the others. Feelings of unreality ("Why does it hit me when I am normally so healthy"), despair, sadness and disappointment were common ("It was a terrible shock for me. I cried, could not leave the clinic but could not stay either").

Five women reported that they had been unbalanced for some time after the abortion but had recovered by 6 months. The other 5 women reported that they had still not recovered. The time from the abortion to the interview in this latter group varied between 9 and 34 months. These 5 women were all strongly emotionally affected when talking about the abortion. In addition, one other patient, who said that she had got over the crisis, showed signs of mental unbalance. Three cried openly. Thus, more than half of

the women (6/10) were still depressed at the time of the interview. One woman reported that she had nightmares thinking that she was in the operating theatre.

The majority (7/10) of the women still thought about how old their child would have been. One patient felt somebody was missing in the family. Another said: "I feel emptiness, I miss her, I mourn her every time I think about her, I try to see her in my mind".

After the abortion all women were offered the possibility to see the fetus. Five (5/10) agreed to this and all 5 expressed a feeling of relief, and of assurance that it had been right to make a legal abortion. The women who chose not to see the fetus, did not regret their decision at a later stage.

Only 2 women admitted that they had subsequently considered they might have been wrong in having the abortion. One said: "Perhaps the child would have survived, perhaps an operation might have been possible?".

Four women had subsequently given birth to healthy children. At the time of the interview, another 2 were more than 17 weeks pregnant, and 2 planned to become pregnant. Only 2 women did not wish a further pregnancy: one had been sterilized in connection with the abortion carried out by hysterotomy, the other already had 3 children.

The 6 women who became pregnant again considered it important to have ultrasound scanning in the 17th week of gestation; 5 also wanted an amniocentesis.

To the question: "Has the fact that you carried a malformed fetus had any influence on your self-esteem?", 3 women (3/10) answered that their self-esteem had been damaged; they still had feelings of guilt and inferiority: "I had a glass of alcohol early in the pregnancy or could it have been the fact that I work with a data

screen that damaged the child?". Seven women had no such feelings; 3 of these had subsequently given birth to a healthy child, and 1 was more than 17 weeks pregnant. Thus, the birth of a healthy child may help to reestablish self-esteem: "I feel better now since we have a daughter".

In the present study there was no indication that the partner relationship suffered from the diagnosis of the malformed fetus. In contrast, 7 reported an improvement: "We have come through a crisis together". Only 1 woman said that her relationship with her partner had deteriorated: "We have difficulties talking to each other; we have such different feelings about the abortion".

Discussion

Obviously, our sample is small, mainly due to the low incidence of fetal malformations. As we considered it important to have a homogeneous group, patients referred from other hospitals were not included. Further more, the aim of the study was not epidemiologic, but to improve understanding of how these patients feel and react in order to be able to offer better emotional support.

Generally, the women were satisfied with the treatment they had received at the ultrasound department. This satisfaction may result from not wishing to criticize a department where the interviewer (CJ) was working. Another reason might be that they have no comparative experience.

The results show that patients having an abortion because of fetal malformation experience a severe psychological trauma, particularly a threat to their individual reproductive capacity. The reaction may be long-lasting. In the present study, half of the patients had not got over the crisis. In some cases, selective abortion will probably cause strong emotional reactions whenever the woman is reminded of it.

Even if only a few patients expressed a need for more help, it is reasonable to conclude that more active psychological support might have been valuable for most of the patients (13, 14). The women require both detailed information and good emotional support. Similar conclusions have been drawn in previous studies, which, however, do not deal with ultrasound screening (15, 16).

It may be important to realize that the time of expected childbirth, usually 6 months after the abortion both in the present study and in other studies (17), may be an particularly difficult period for the patients, and that psychosomatic symptoms may appear at this time. A medical appointment with the same doctor may well be advisable.

The crisis reactions are comparable to those described following perinatal death (4, 18). Hence, the responsibility of the doctor to provide supportive contact to parents with a fetal loss must be as extensive as to parents experiencing a perinatal loss. It may even be the case that the parent's feelings of guilt are more accentuated when they have decided to have a selective legal abortion than in cases of perinatal death. Furthermore, in many ways their relationship to the medical establishment may be characterized by ambivalent feelings. At the same time as expecting themselves to be grateful to those people for discovering the fetal malformation, they also feel grief and aggression for having been bereaved of their child. Feelings of grief, guilt and anger may be difficult to communicate to people to whom one is expecting to be grateful. This makes the position of the obstetrician somewhat precarious. However, we consider it important that the person who has been responsible for the diagnosis of the fetal malformation should also be responsible for giving emotional support to the parents.

As with perinatal death, it seems important that the patients see their fetus. Half of our patients did so and expressed a feeling of relief. Seeing the fetus provided them with a concrete experience which could function as a substantial background for their grief.

A major problem is whether there is any advantage by diagnosing fetal malformation already in the second trimester. Some of the pregnancies in this study would probably have ended in a spontaneous abortion. The routine ultrasound scans in the 17th and 32nd week of gestation were not primarily carried out in order to diagnose fetal malformations. However, in the present study, the women considered the early diagnosis and the induced abortion valuable. For most of them it was beneficial to avoid a delivery resulting in a malformed and not in a healthy child (19).

It has become increasingly common that fetal malformations are discovered antenatally. This is partly due to investigations with the aim of excluding fetal malformations and partly due to new methods of pregnancy surveillance. The present study, like previous ones (15, 16, 17, 20), clearly indicates that obstetricians using these methods, must be prepared to handle the subsequent psychological reactions. In many cases it may be sufficient to be aware of the crisis experienced by the patients and to provide supportive contact with the main purpose to help the patients to react to and get over the trauma. Some patients may be mentally disturbed to such a degree that long-term psychotherapy is required. This may be particularly true when the woman has already previously had traumatic experience within the sphere of reproduction and sexuality.

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VI

Diagnosis of fetal malformation in the 32nd week of gestation. A psychological challenge to the woman and the doctor

C. Jørgensen, N. Uddenberg & I. Ursing

Abstract

During a three year period 15 cases of fetal malformation were diagnosed by routine ultrasound scanning in the 32nd week of gestation. Fourteen women were interviewed in order to find out how this diagnosis influenced the remaining part of the pregnancy, the delivery and the future reproductive life. In one case intrauterine fetal death occurred in gestational week 36, this woman was excluded from the study. The women were divided into two groups according to the type of malformation of the fetus. In one group the children would probably have some permanent injury, in the other the children could, after adequate treatment, be expected to be healthy. All the women were emotionally unbalanced during the rest of the pregnancy and had great difficulties in forming a realistic picture of their child. At the time of the interview all women had adapted to their situation. Eleven women were informed of the fetal malformation directly after the ultrasound examination whereas three were not informed before the delivery. All 14 women considered it not acceptable not being informed as soon as the malformation had been diagnosed. Discovering a fetal malformation, late in pregnancy commits the woman to the fact that she has to give birth to a malformed child. The situation requires the obstetrician to be prepared to support the parents by offering repeated contacts and opportunities to discuss the situation.

Introduction

Ultrasound scannings of pregnant women in the 2nd and 3rd trimester have become indispensable for the obstetrician in situations of pregnancy complications. Even used routinely in all pregnancies ultrasound scannings have got an increased application. These scannings may influence the women's behavior on given health advice (1) as well as her anxiety level (2).

Ultrasound screening has been used at the University Hospital of Lund since 1st October 1980. All pregnant women in the district of Lund have been offered two examinations, i.e. in the 17th and 32nd week of gestation, respectively. At the 32nd week the primary intention is to control the growth of the fetus although information about fetal malformation is also obtained (3, 4). The psychological reactions when a fetal malformation is discovered at the 17th week of gestation have previously been described (5).

When a fetal malformation is discovered in the 32nd week of gestation the parents have to accept to continue the pregnancy and to give birth to a child with some kind of malformation. The mother's reaction may be influenced by the degree of the child's handicap. Some congenital malformations are surgically correctable resulting in a healthy child. Others may be influenced to some extent by treatment but some kind of permanent disability will remain.

Living with a handicapped child the parents have both to establish a loving relation to their child and to cope with concrete practical problems (6, 7). The possibility for survival of the child will in some cases of fetal malformation be very uncertain and the mother may feel the threat of losing her baby (8, 9). Furthermore, the mother will be confronted with the fact that she was not able to give birth to a healthy child. This may influence her self-esteem and result in feelings of guilt (10).

The aim of the present study has been to increase the knowledge of the woman's experience of the given information of fetal malformation (diagnosed in the 32nd week of gestation), her reactions during the remaining part of the pregnancy, the delivery and the following years. Such knowledge may help the obstetrician to offer a better emotional support.

Patient sample

During a three year period a total of 8422 women were examined twice during their pregnancy with a routine ultrasound scan. Fetal malformations were found in 15 pregnancies in the 32nd week of gestation. All these women had been examined in the 17th week of gestation with normal results. One fetus died in utero in the 36th week of gestation. On ultrasound examination and later autopsy the infant was found to have an extremely dilated urinary bladder of uncertain genesis and died from unknown reason. This case was not included in the study.

Table I shows diagnosis, time and method of delivery and the condition of the child at the time of the interview according to the pediatrician and the mother, respectively. The children were divided into two groups depending on the character of the malformation. One group would probably have sequelae (S, cases no. 1-8) the other would have no sequelae after treatment (NS, cases no. 9-14). All 14 women were later contacted for an interview. The time from the birth of the child to the interview varied between 3 2/12 years and 7 months with a mean of 1 7/12 year. Thirteen of the women lived with the same partner at the time of the diagnosis as at the time of the interview. One had separated and lived alone with her child. The age of the women at the time of the interview varied between 22 years and 35 years with a mean of 29 4/12 years.

Four of the women had a university degree, four college education and six basic school. At the time of diagnosis six patients were primigravida, three had had a legal abortion and five had children previously. None of the women had received any treatment for infertility.

Table I. Delivery week and method, diagnosis and the condition according to the pediatrician and the mother.

Case No.	Delivery week	method	Diagnosis	Pediatrician	Follow-up	Mother
1	39	S	Lumbosacral myelomeningocele	Lower leg flaccid paresis. Bladder paresis. Shunt op. Normal psychomotor development.		Slight handicap
2	42	V	Lumbosacral myelomeningocele	Bladder paresis. Shunt op. Normal psychomotor development.		Slight handicap
3	38	V	Hydrocephalus. Coxsackie-virus	Slight spastic diplegia. Moderate psychomotor retardation.		Slightly retarded one year
4	37	S	Meningoencephalocele	Epilepsy. No paresis. Shunt op. Normal psychomotor development.		Healthy
5	37	V	Thoracolumbal myelomeningocele	Lower body flaccid paresis. Shunt op. Normal psychomotor development.		Moderate handicap
6	38	S	Dandy-Walker syndrome	No paresis. Shunt op. Normal psychomotor development.		Healthy
7	33	S	Stenosis aqueductus cerebri	No paresis. Shunt op. Slight psychomotor retardation.		Slightly retarded
8	39	V	Phocomelia extrem. inf. dx.	Normal psychomotor development. Artificial limb planned.		Moderate handicap
9	43	S	Hydronephrosis left kidney	Ureteroneocystostomy Healthy		Healthy
10	38	S	Gastroschisis	Primary skin closure. Secondary hernioplasty. Healthy.		Healthy
11	41	V	Right polycystic kidney Colonic atresia	Operation of atresia. Nephrectomy. Slight hypertonion.		Healthy
12	33	S	Duodenal atresia	Operation of atresia. Healthy.		Healthy
13	34	S	Hydronephrosis right kidney	Pyeloplasty. Nephrectomy. Healthy.		Healthy
14	38	V	Right polycystic kidney	Nephrectomy planned.		Slightly injured

Diagnostic procedure.

The ultrasound screening was performed by specially trained midwives. When a fetal malformation was suspected, the patient was referred to an obstetrician with special experience of ultrasound investigations. The obstetrician confirmed the diagnosis and informed the patient. This took place as soon as possible after the first suspicion of fetal malformation. The patient was followed with ultrasound examinations, during the remaining part of the pregnancy, at an interval of one to three weeks depending on the diagnosis. The findings were discussed with the parents, and in most cases they also met specialists in paediatric surgery or neurosurgery. A visit to the Neonatal Intensive Care Unit was included in the program. The method of delivery was discussed with the woman and it was electively planned in those cases where the condition of the infant or the mother needed special consideration. Thus, the parents were actively taking part in the decisions. If possible, the infant and the mother were together in the puerperal unit, but most of these infants needed special observation in the Neonatal Intensive Care Unit. This unit is situated next door to the puerperal unit allowing an intimate contact between mother and child.

In the sample 11 of the 14 women were handled as described above. Three women had however, no information of the fetal malformation during their pregnancy; for various reasons the obstetrician preferred not to inform the woman.

Methods

In January and February 1984 the 14 women with diagnosed fetal malformation were contacted by a letter informing of the present investigation. Later they were contacted by telephone and a time for an interview was arranged. All 14 women accepted to participate.

The interview was loosely semi-structural, i.e.

- 1) Questions designed in advance were asked in a certain order.
- 2) The woman was allowed to elaborate her answers freely.
- 3) The interviewer wrote down the essential of the women's answers, which could later be categorized.

The interview contained questions of the women's reactions when being informed of the fetal malformation, the experience of the remaining part of the pregnancy, delivery and puerperal period, and how the birth of the child had influenced the lives of the women.

Seven of the 14 interviews took place at the Department of Obstetrics and Gynecology, 6 at the home of the women and 1 at the woman's place of work. The time for an interview was scheduled to 45 minutes but the actual time spent was between 3 hours and 45 minutes with a mean of 1 1/2 hour. The atmosphere during all the interviews was friendly and informal.

Further information was obtained from the hospital files of the women and children.

For statistical analysis 2x2 tables and Fishers exact test (two tailed) were used.

Results

The women's experience of given information.

Three women had no information of the diagnosis during the pregnancy (vide infra). The remaining 11 had already during their pregnancy got detailed information of diagnosis, possible treatment and prognosis for the child. Nine of these women reported that they had found the information sufficient, 2 had felt a need for further discussion. Six women (6/11) mentioned that the information given

by the pediatrician, the pediatric surgeon or the neurosurgeon was of great importance: "It proved that someone cared about us".

Seven women (7/11) considered that they had understood the implications of the malformation. In spite of information, accurate from a medical point of view, 4 women had, however, found it difficult to realize the consequences for the child: "We got all possible information but could not grasp that she was so injured".

Questioned if after the information they had experienced the fetus as "kind of a monster", five women (5/11) denied such feelings. Still, some of these women may have had thoughts in this direction. For instance one woman said: "It would have been wrong to think so". Six women, reported "monster feelings" or feelings that their child had disappeared: "I had a big belly without contents, the womb was empty; there was no baby any more". Another woman said: "I thought about the head of the baby which was too large for the body". Another could not give up the thought that the baby could have other, still not detected malformations. Only four women (4/11), reported that they had been able to think of their child in a reasonably realistic way and could still imagine that they were going to have a baby. There was no association between the woman's picture of the child and the type of malformation (Table II). Obviously, detailed information does not preclude unrealistic fantasies.

The three women who had no information during pregnancy, had repeated ultrasound examinations, motivated by control of the fetal growth. All three had suspected that something was wrong, had wanted an explanation, but had not had the courage to ask. At the interview they reported that they had felt deceived and that their confidence in the obstetrician had been severely disturbed.

The women's experience of the remaining part of the pregnancy.

The information that the child was malformed naturally aroused intensive emotions. All the women described the remaining part of the pregnancy as dreadfully straining. Some (4/11) wanted to be delivered as soon as possible in order to "put it all to an end". Others (3/11), on the contrary, wanted to postpone the delivery not to be confronted with the child.

The majority of the women (9/11) reported that they had received strong emotional support from their husbands.

Experience of delivery and puerperium.

Eight (8/14) women had a Cesarean section performed, one of which was not planned. Fetal malformation was the only indication for the Cesarean section in three cases. In four cases there were other obstetrical indications. One woman had a Cesarean section for psychological reasons. Six had a vaginal delivery; only one of these reported that she would have preferred a Cesarean section.

From an obstetrical point of view all women had a normal puerperium. Extra consumption of analgesic and sedatives was not observed. Nearly all (13/14), reported that they had felt emotionally unbalanced and depressed during the puerperium: "I felt terrible, wanted to hide under the blanket".

Most women (4/5) who had had strong feelings that the child was a "monster", felt relief when reality turned out to be less frightening than fantasies. In one case (no. 7) the relief was strong enough to prevent emotional unbalance.

For some women there was a predominant fear that the baby should die: "I was afraid of being too attached to the child and let my feelings free if he should not survive". During the first days

after delivery two women experienced difficulties to look at and to touch their baby and emotionally to accept their motherhood (no. 1 and 11).

The time from delivery to interview.

The period of emotional unbalance reported by the group of women with S-children varied between a few days to 2 years and in the group of women with NS-children between 1 and 7 months.

The duration of emotional unbalance, however, depended to a great extent upon the medical treatment of the child. When the child could be definitely cured by an operation e.g. hydronephrosis and intestinal atresia, the mother recovered.

The mothers with S-children also reported a concrete experience to be important for the change towards better emotional balance: "When I saw him laugh and move". The mothers of infants with CNS-malformations (cases no. 1-7 table I) did not report longer periods of emotional unbalance than the others (mean 5 1/2 months).

At the time of the interview all women considered themselves mentally well. None showed signs of emotional disturbance when discussing the period when the fetal malformation was discovered.

For seven women the relationship to their husband was uninfluenced, whereas three found it improved: "We have worked it out together, we are more attached to each other, we have learnt to appreciate what we have". Four women reported a deteriorated relationship: "We cannot talk together, we are too anxious and worried", another said: "He does not feel as responsible as I do". Only one woman had been divorced. There was no significant correlation between the type of malformation and the women's relation to the partner (Table II).

Attitude towards the child at the time of the interview.

At the interview, eight of the women did not regard their child as handicapped and expected a normal development, including two whose children had shunt-operated congenital hydrocephalus (cases no. 4 and 6).

On the other hand one woman in the group of NS-children answered: "I cannot treat him as a healthy child. I am afraid he shall cry too much so that the cyst will burst". This child had a unilateral polycystic kidney and was expected to be cured by an operation (case no. 14). Thus, the mother's opinion about her child's health was not always in accordance with that of the pediatrician (Table II).

Most of the mothers with S-children (6/8) reported that if the malformation had been discovered during early pregnancy they would have preferred to terminate the pregnancy. None of the mothers of NS-children would ever have considered an abortion.

In accordance with this nearly all (7/8) of the mothers with S-children would accept an offer of an amniocentesis in case of a future pregnancy.

Attitude to future pregnancies at the interview.

Nine of the 14 women had no children previously. At the time of the interview two of these women had given birth to another child and one was pregnant. The remaining 6 wanted children sooner or later. Four of the 5 women who had children previously had no plans for more and one had given birth to another child.

None of the women stated that she dared not have another child because of the risk of malformation. Probably, the birth of the malformed child did not alter radically the women's attitude towards future pregnancies.

Table II Comparisons of the women's answers, between those who had S-children and those with NS-Children.

		S-children	NS-children	Fishers exact test
Had a realistic picture of the child	yes	4	4	p = 0,70
	no	4	2	
Had need of more contact	yes	4	2	p = 0,70
	no	4	4	
Better or unchanged relation to husband	yes	6	4	p = 0,80
	no	2	2	
Regard the child as healthy.	yes	2	5	p = 0,09
	no	6	1	
Expect a normal development.	yes	2	4	p = 0,28
	no	6	2	
Would have abortion if early diagnosis	yes	6	0	p = 0,02
	no	2	6	
Want amniocentesis in future pregnancy	yes	7	0	p = 0,005
	no	1	6	

Discussion and Conclusions

The present study is based upon the first 14 cases of fetal malformation diagnosed accidentally by routine ultrasound scanning in the 32nd week of gestation and delivered at the University Hospital of Lund, Sweden. Our study presents some first impressions of the psychological problems connected with ultrasound sceening in the third trimester, in order to increase the understanding and improve the clinician's management of these cases. The design of the study implied that no standardized follow-up time could be used. Evaluating the answers it is important to realize the possibility of selective recall and the fact that the interviewer (CJ) was one of the doctors responsible for the diagnosis and obstetrical management.

Information

The results of the study do not support the opinion that it should be considerate to the women to postpone information about fetal injury until the baby has been born. Those women in the present study who got no information during the pregnancy, all suspected something to be wrong and felt deceived and disappointed afterwards. All the women stated that it was important to get information as soon as possible, a finding which is in accordance with observations of mothers who have born a malformed child (11, 12). Probably, the postponing of information was the doctor's way of avoiding an unpleasant situation.

In spite of detailed information, the women obviously had great difficulties in obtaining a realistic picture of their child. Even women who were informed that their child had a relatively small and correctable malformation, might imagine that the baby was "kind of a monster" (6, 7, 12). It is well known, that an individual's capacity to assimilate traumatic information is limited (7, 10, 13). Repeated contacts with the obstetrician are important both as a form of emotional support and as an opportunity to get information about the child.

Obstetrical management

The study shows that many malformed fetuses may be delivered vaginally. Most of the women, both those who had a Cesarean section and those who had a vaginal delivery, were content with the method chosen. One reason for this could be their active part in the process of decision.

Several women reacted to the information that the child was malformed with a wish to bring the pregnancy to an end, in order to get rid of the malformed baby. The obstetrician, in contrast, wanted to postpone the delivery in order to increase the child's possibilities of survival. The woman, torn between horror and hope, may

have difficulties in accepting the doctor's advice. Her reluctance may be connected with "forbidden wishes" that the child should die (6, 7). The woman's natural ambivalence towards her malformed child increases her need of an open discussion with the obstetrician.

The time to the follow-up interview

During the puerperium the mother's feelings of ambivalence remain. She both wants to take care of her child and to avoid to be confronted with the malformation. It is important that the staff at the puerperal ward understands her dilemma. Generally, the women were emotionally unbalanced during the puerperium. Mothers of children with less severe malformations described equally strong reactions as mothers of children with severe injuries.

Nearly all children with congenital malformations need care at the intensive care unit, which means an early separation. The prognosis for the child's survival may in some cases be uncertain implying that the mother may experience a threat to lose her child which may seriously influence the emotional attachment (7, 9). In addition the ambivalence of the mother may result in inadequate mother-child bonding. It is important to keep this in mind and to give the mother opportunities to see and care for her child (12, 14, 15, 17).

At the time of the interview, none of the women showed signs of mental unbalance. Even if a few women still had unrealistic fantasies (positive or negative) about the child, most of them seemed to have resolved the crisis remarkably well. This may partly be due to psychological defence mechanisms (7, 18). However, these women have also been forced to solve the practical problems and emotional conflicts connected with having an injured child, which may increase their self-esteem. In one way or other they seemed to have accepted their situation, although some of the mothers with severely handicapped children expressed ambivalence towards the child, admitting that they would have had an abortion if possible.

Obviously, the parents have a need to continue their contacts with the obstetrician even after the birth of the child. They need both to discuss their experiences of bearing a malformed child and to be counselled in case of future pregnancies.

Ultrasound scanning is a practical tool to solve a number of obstetrical problems. However, the obstetrician using this method must also be prepared to take care of the emotional conflicts connected with unexpected discovery of fetal malformation. The present study shows the importance of being open with the mother, listen to her fantasies and understand her ambivalence towards her baby.

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